Siblings of pediatric cancer patients: psychological adjustment and interventions

Thesis
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Abstract

Over three research studies, this thesis explores psychological adjustment and interventional approaches in siblings of pediatric cancer patients.

Following a *general introduction*, the *first study* reviews the existing reports of evaluated psychological interventions with siblings of cancer patients. Research papers and dissertations included in the review were selected on the basis of pre-defined criteria. Data from fourteen studies, representing eleven different sibling interventions, met criteria for inclusion. Seven groups, three camps, and one individualized intervention for siblings were found. Empirical evidence on the effectiveness of these interventions was analyzed by summarizing outcome variables and calculating effect sizes, whenever possible. Furthermore, the objectives and settings of the interventions were described. Evaluation findings revealed significant improvements in siblings' depression symptomatology, medical knowledge, and health-related quality of life (HRQoL). Findings were inconsistent with regards to anxiety, behavioral problems, social adjustment, self-esteem, and posttraumatic stress symptoms. Depending on the outcome variables, small to large effect sizes were identified. Satisfaction with the intervention was high among both siblings and parents.

In the *second study*, the findings of a qualitative study on siblings' experiences in the first half-year after the cancer diagnosis, are presented. In semi-structured interviews, seven siblings of pediatric cancer patients were asked about their experiences in the hospital, in school, within their family, with peers, and with the ill child. Content analysis was used to extract important themes from the interviews. Twenty-two categories of sibling experiences were identified from the data. Across all areas of life, siblings reported difficulties, such as the absence of parents, dealing with the ill child's or other patients' suffering and appearance, or impaired school achievement. However, siblings also mentioned important resources, like peer relationships, helpful coping strategies, and increased family cohesion.

The *third study* evaluates the effectiveness of a two-session psychological intervention for siblings of newly diagnosed pediatric cancer patients. Thirty children (age 6–17 years) were randomly assigned either to an intervention or to a control group. The manualized intervention provided to siblings within the first 2 months after the cancer diagnosis included medical information, promotion of coping skills, and a psycho-educational booklet for parents. At 4 to 6 weeks, 4 months, and 7 months after the diagnosis, siblings and their parents completed measures of psychological adjustment (anxiety, depression, posttraumatic stress), social support, health-related quality of life, coping, and medical knowledge. At follow-up, siblings in the intervention group exhibited better psychological well-being and a greater degree of medical knowledge, and reported receiving social support from more people. However, the intervention had no effects on any other adjustment measure.

Finally, a *general discussion* integrates and discusses the findings of all three studies.

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Tables and Figures

I. General Introduction

When a child's diagnosis is cancer, sisters and brothers are usually – and naturally – not at the center of attention. Nevertheless, they should be attended to. In this thesis, the focus is exactly on this population. The aims of the present doctoral thesis were to examine psychological adjustment and evaluate interventions for siblings of pediatric cancer patients. Prior to a discussion of the current state of intervention research in Chapter II, the presentation of results of a qualitative pre-study in Chapter III, the evaluation of our self-developed intervention in Chapter IV, and an overall summary and discussion in Chapter V, the present introductory chapter provides some background information on the topic concerned. It includes an overview of medical basics in pediatric oncology, introduces the field of pediatric psycho-oncology, and gives a summary of the consequences of cancer for the family and siblings, specifically. Further background information on the present research project, with a description of objectives and methods, is given.

1. Pediatric Cancer

1.1 Epidemiology

Cancer is predominantly a disease of aging and its occurrence in children is fortunately rare (Heath & Ross, 2010). Despite this rarity, cancer remains the second most common cause of death in childhood in developed countries, after injures, and a significant public health problem (Jemal et al., 2006).

Based on data from the Swiss Childhood Cancer Registry (SCCR), a national hospital-based cancer registry with very high coverage rate, the age-adjusted incidence rate for pediatric cancer in Switzerland for the decade 1995 to 2004 was 14.1 per 100 000, with an average of 174 new cases of cancer diagnosed in children under age 15 years each year (Michel et al., 2008).

The types of cancer seen in children generally are different from those encountered in adults. In children, cancer typically develops in tissues and organs that grow most rapidly during embryogenesis and the postnatal period (Izraeli & Rechavi, 2004). Conversely, typical adult malignancies arise in epithelial cells covering the surface of ducts and body cavities that are exposed for prolonged periods of time to a large variety of environmental carcinogens (Izraeli & Rechavi, 2004). While in adults about 80 % of cancer cases pertain to the respiratory, gastrointestinal and reproductive organs, less than 5 % of cancers in children are manifested in these organs (Imbach & Kühne, 2006).

While adult cancers are frequently the result of living style and environmental causes, there is much less evidence for the influence of environmental or behavioral factors on the etiology of cancer in childhood (Heath & Ross, 2010). Most pediatric cancers cannot be prevented, are not preceded by obvious premalignant lesions, and are not amenable to early diagnosis, a

fact most relevant when dealing with parents of patients, who typically are overwhelmed by guilt and self-blame (Izraeli & Rechavi, 2004).

Childhood cancers are extremely diverse, both in origin and distribution (Heath & Ross, 2010). As seen in Figure 1, one third of childhood cancers diagnosed in Swiss children between 1976 and 2007 were leukemias (35%), followed by central nervous system (CNS) neoplasms (brain tumors), and lymphomas, comprising 18% and 14% of all pediatric tumors, respectively (Kuehni et al., 2009). These three major diagnostic groups will be explained in more detail below.

The most common pediatric cancer diagnosis is *leukemia*. It is a cancer of the bone marrow or of the white blood cells. Symptoms of leukemia in pediatric patients include persistent fevers, infections, bruising, pain in the bones and joints, and enlargement of the lymph nodes (Granowetter, 1994). In acute lymphoblastic leukemia (ALL), abnormal lymphoblasts aggregate in the bone marrow, resulting in the failure of bone marrow production (Brown, 2006). The normal lymphoid system grows rapidly during embryonic development and early childhood, and acquires the capability of mounting specific immune responses against an enormous variety of foreign antigens (Izraeli & Rechavi, 2004). Lymphoid cells possess an unusual type of genetic instability, allowing for the diversification of various immune receptors; but this also predisposes them to rare genetic accidents, some of which lead to acute leukemia (Izraeli & Rechavi, 2004). Acute lymphoblastic leukemia accounts for about 75% and acute myeloid leukemia (AML) for about 20% of pediatric leukemias (Pui, 1999). AML is a cancer of the blood-forming tissues that do not include the lymphoblasts (Granowetter, 1994).

Malignant *brain tumors* are the second most common type of cancer observed in childhood. These tumors are heterogeneous in nature, and symptoms vary in accordance with the site and size of the tumor. Typical symptoms include headaches, vomiting, double vision, difficulties with balance, and cognitive or neurological deficits (Brown, 2006). The prognosis for childhood brain tumors is also largely associated with the site of the tumor, the amount of CNS infiltration, and the specific histological subtype of the tumor. Despite advances in treatment, brain tumors remain the leading cause of death in childhood cancer patients (Heath & Ross, 2010) and many survivors face physical, psychological, social, and intellectual challenges directly related to their treatment (Turner, 2009).

Lymphomas are a group of malignancies in which the cancer cells originate from the lymphoid system. Lymphomas include non-Hodgkin's lymphomas (NHLs) and Hodgkin's lymphomas. Hodgkin's lymphoma typically has a slower onset and more orderly progression than NHLs (Granowetter, 1994). Common sites for NHLs include the head and neck, the abdomen, and the chest. Symptoms and signs include enlarged lymph nodes, fevers, weight loss, lethargy, and abdominal masses (Brown, 2006).

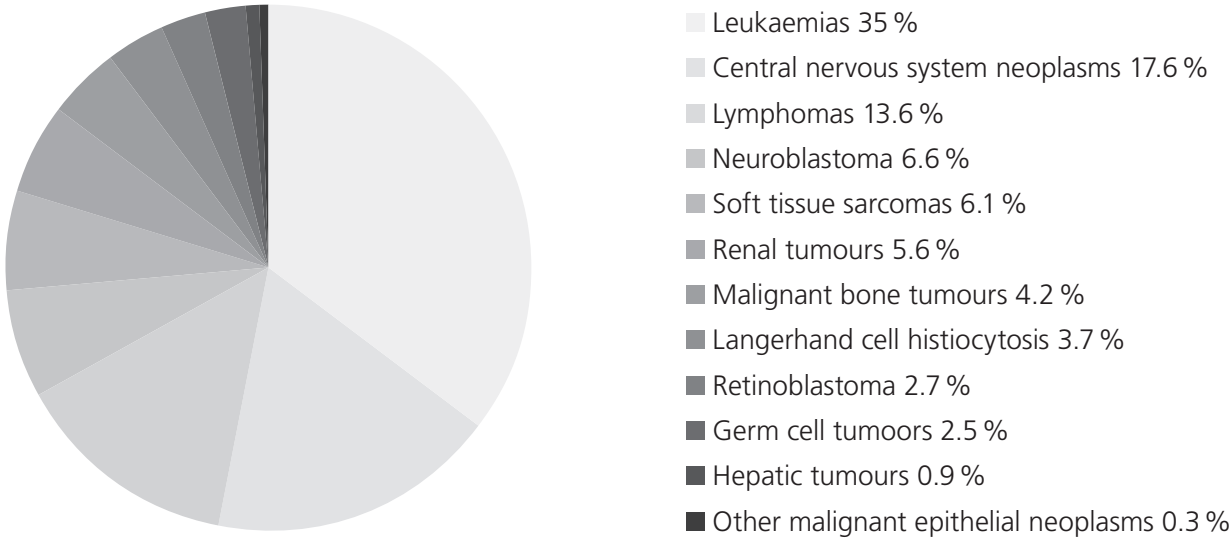

Figure 1. *Diagnostic groups aged 0–14 years at diagnosis, Swiss residents, years 1976–2007 (Swiss Childhood Cancer Registry, 2009).*

1.2 Treatment and Prognosis

Treatment of pediatric cancer is a story of success in modern medicine. Mortality rates for all malignant childhood cancers combined declined by more than 50% between 1975 and 2006 (Smith et al., 2010), such that survival rates in childhood cancer are now reaching 75% to 80% (Gatta et al., 2009). Still, improved prognosis and survival require prolonged, complicated, and intensive treatment protocols (Long & Marsland, 2011). Four primary modalities of therapy are available for pediatric cancer: chemotherapy, radiation therapy, surgery, and bone marrow transplantation. It is not uncommon for a single patient to receive a combination, consisting of two, three, or even all four of these modalities.

Chemotherapy is the main component of most cancer treatment protocols, as it can be administered systematically to work at the primary tumor site as well as throughout the body (Granowetter, 1994). Chemotherapeutic agents typically exert their effect by targeting the unduly rapid growth of cancer cells, and are administered through the veins, intramuscularly, or into the spinal fluid (Brown, 2006). There are a number of adverse side effects associated with chemotherapy. Most of these adverse effects are generally short term, such as nausea, vomiting, hair loss, diminished appetite, mouth sores, general malaise, and low blood counts that make the child susceptible to infection, and can in many cases be alleviated by medication. Of greater concern are the long-term adverse side effects of the chemotherapies, which can affect the heart, kidneys, and liver (Brown, 2006). Although children tolerate the acute toxicities of chemotherapy better than adults, growing children are more vulnerable to the delayed effects of cancer therapy, like its effects on growth, the endocrine system, fertility, the myocardium, and neuropsychological functioning (Izraeli & Rechavi, 2004).

Radiation therapy uses high-energy radiation to shrink tumors and destroy cancer cells by damaging their DNA (Lawrence, Ten Hake, & Giaccia, 2008). This therapeutic approach is

often used for a tumor when surgery and chemotherapy cannot completely remove it, and it is typically administered over a series of days or weeks (Brown, 2006). The treatment sessions usually take only a few minutes, but require that the child lie very still. For smaller children, this is difficult, such that sedation or anesthesia may be needed (Granowetter, 1994). Radiation therapy itself is painless, but there are a number of potential adverse side effects. These include lethargy, loss of appetite, and irritation of the skin or mucous membranes in areas where the radiation has been directed (Brown, 2006). If radiation therapy is administered to the CNS, one of the more serious adverse effects is a decrease in white matter in the brain, which may result in significant learning impairments and, in some cases, in mental retardation (Mulhern, Merchant, Gajjar, Reddick, & Kun, 2004).

Surgery is a local therapy to remove the malignant tissue and an essential part of the treatment for many solid tumors. Tissue around the tumor and nearby lymph nodes may also be removed during the operation. Sometimes, radiation or chemotherapy is used first to shrink the tumor before it is removed (NCI, 2003).

Bone marrow transplantation refers to the transplantation of the marrow of the bone or the cells that produce blood-forming tissue. Is a treatment modality particularly used for children diagnosed with leukemia or solid tumors (Granowetter, 1994). Bone marrow transplantation starts with intensive chemotherapy, which almost completely wipes out all bone marrow function. Subsequently, new bone morrow is administered to the patient intravenously. The new bone marrow may be either that which was collected from the patient during a prior period of remission (autologous transplant) or a transplant from a healthy donor. Until bone marrow transplantation is completed, the patient is at high risk for infections and protective isolation is necessary. A major complication associated with bone marrow transplantation is graft-versus-host disease, in which the recipient's immune system responds to the donor's cells as foreign (Brown, 2006).

The length of cancer treatment varies and depends upon the diagnosis; but it usually lasts for several years. As an example, the treatment protocol for childhood ALL consists of 2–3 years of therapy, utilizing up to 10 chemotherapeutic drugs given in various combinations (Izraeli & Rechavi, 2004). Intensive remission induction and consolidation therapies last for up to half a year and require frequent hospitalizations for the administration of drugs or for combating infectious complications of chemotherapy. This intensive period is followed by prolonged, less-intensive maintenance therapy, during which hospitalizations are less common and children generally can attend school again (Izraeli & Rechavi, 2004). Besides the cancer treatment itself, monitoring treatment response involves further multiple medical procedures, from repeated blood draws to bone marrow aspirations and lumbar punctures (Long & Marsland, 2011).

2. Pediatric psycho-oncology

Along with the success of therapeutic means in pediatric oncology and important changes in information and communication practices, the discipline of pediatric psycho-oncology devel-

oped and became established in clinical practice and research. Historically, during the 1950s, no effective chemotherapy was available for pediatric cancer patients, so that the focus could only be directed towards caring for the dying child and preparing the family for that child's inevitable death (Eiser, 1994). Psychosocial research in pediatric psycho-oncology began in the 1960s and was continued in the 1970s via studies that were predominately observational, examining parental mourning processes and psychological reactions to the death of their child (Patenaude & Kupst, 2005). The research subjects largely were parents, since the children themselves were excluded from discussions about their disease and usually were not offered information about the probable diagnosis or the expected course of their illness (Eiser, 1994). Through the 1970s, effective therapies were developed and psychological research shifted its focus from "dying" to "living with a life-threatening condition" (Eiser, 1994). As survival rates started to improve, attitudes towards open communication about diagnosis and prognosis with pediatric cancer patients changed notably. These changes in communication were influenced by studies showing that ill children were not deceived by the avoidance of discussion of their disease, and that they were able to understand much more about their illness and prognosis than was commonly assumed (Spinetta, 1974). In order not to leave the children alone to cope with their fears, open communication about cancer began to be emphasized by health care professionals. Professionals started to speak directly to the patients about their illness and they encouraged their parents to do the same (Patenaude & Kupst, 2005).

The change towards open communication eventually resulted in the inclusion of mental health professionals into treatment teams for children with cancer. Psychologists, psychiatrists, and social workers became more and more involved in pediatric cancer services. These experts took over the function of helping families to explore and cope with the challenging issues of life-threatening illness and imminent death; but they also fostered the awareness that the diagnosis has an impact not only on the ill child, but on the whole family system, including siblings (Patenaude & Kupst, 2005). Analogously, psychosocial research in the field increased. Since children who were cured of their cancer could expect to live many more productive years, quality of life and the late effects of cancer and its treatment have become a major focus of recent pediatric oncology, and are an important issue in the field of psycho-oncology research (Izraeli & Rechavi, 2004).

3. Pediatric cancer and family functioning

From birth to late adolescence, the family unit is primarily responsible for the nurturance and protection of a child (Gass, Jenkins, & Dunn, 2007). Parents or primary caregivers generally are a secure source of support and comfort when children experience distress, anxiety or fear (Bowlby, 1982). Childhood cancer is a highly stressful experience that challenges the whole family system and may even disrupt it (Alderfer & Kazak, 2006).

Such family changes in the context of childhood cancer have recently been examined in a comprehensive review of quantitative and qualitative publications (Long & Marsland, 2011).

Families must cope with the fact that the course of cancer is unpredictable and uncontrollable, and with the possibility that their child will not respond to treatment (Long & Marsland, 2011). Thus embedded into a family system, children with cancer and siblings are very likely to affect and be affected by the family's reaction to the diagnosis of childhood cancer (Long & Marsland, 2011). Concerning family functioning, the authors of the review found that mean levels of family functioning do not differ from norm samples, though subgroups of families show ongoing family impairment. Qualitative studies have revealed that the roles and responsibilities of mothers and fathers shift in response to cancer, leaving mothers in general with the caretaking responsibilities of the ill child, while fathers attend to the household, work, and sibling care. It was shown that these changes can contribute to disruptions in daily routines, employment changes, and economic problems, and seem to appear most pronounced during diagnosis and early treatment. Concerning marital quality, a substantial subset of parents reported persistent marital problems. The tendency to put the needs of the marriage and partner on hold during intense treatment stages and incongruence between partners' coping styles represented challenges for the parents. However, in the reviewed studies, many parents indicated that their spouse is their primary source of support and reported enhanced closeness over time. Finally, parenting in childhood cancer families was investigated in some studies. Here, reports suggested that parenting practices shift to include increased overprotection and indulgence of the ill child, although many aspects of parenting remain largely unexplored.

Most studies assessing family functioning related to sibling outcomes in particular are of a qualitative nature. In summary, the findings of these qualitative studies reveal important family topics relevant to the sibling experience of childhood cancer, including the loss of attention within the family, changes in family roles and relationships, disruptions in normalcy, and a reduced sense of security within the family (Alderfer et al., 2010). Quantitative studies regarding this topic are scant and do not reveal many differences between cancer siblings and the general population, in terms of family environment and relationships (Alderfer et al., 2010). The family as an important domain of siblings' daily life is also discussed in Chapter III.

4. Siblings of children with cancer

Psychological and social consequences for siblings are understandable, given the range of changes and experiences with which they are confronted after their sibling receives a cancer diagnosis. These changes include physical and emotional unavailability of the parents, due to their care of the ill child and their own distress, changes in daily routines that disrupt the sibling's day-to-day functioning, and, not least, their witnessing of physical changes in the ill child and personal worries about their brother or sister (Alderfer et al., 2010).

Most psychological research in pediatric oncology includes either patients alone or patients and mothers, with a lack of attention paid to siblings, fathers, or others in the child's social ecology (Kazak, 2005). This focus of research and literature on the sick child may reflect the

siblings' position in the family during the illness process (Houtzager, Grootenhuis, & Last, 1999). While psychosocial effects of childhood cancer are beginning to be understood for patients and parents, the impact upon healthy siblings is less clear (Alderfer & Noll, 2006). A relatively small, methodologically limited body of work yields frequently mixed results on psychological adjustment in cancer siblings (Long & Marsland, 2011). However, the research on this topic has grown over the past three decades. Since the 1980s, two important review articles have been published summarizing empirical research on the psychological adjustment of siblings of pediatric cancer patients (Alderfer et al., 2010; Houtzager et al., 1999).

4.1 Psychological adjustment of siblings

The results of quantitative studies evaluating psychological adjustment generally have been mixed. Overall, these studies demonstrate that the general *emotional* and *behavioral adjustments* of siblings are within normal limits, with most derivation from normalcy apparent shortly after the diagnosis of cancer has been made (Alderfer et al., 2010).

With regards to *anxiety* and *depression* scores across samples of cancer siblings, the results of studies are inconclusive (Barrera, Chung, Greenberg, & Fleming, 2002; Cohen, Friedrich, Jaworski, Copeland, & Pendergrass, 1994; Dolgin, Somer, Zaidel, & Zaizov, 1997; Houtzager, Grootenhuis, Caron, & Last, 2004; Sahler et al., 1994; Sloper & While, 1996). However, most of these studies identified no elevations in clinical anxiety or depression (Alderfer et al., 2010).

Cancer-related distress in the form of *posttraumatic stress (PTS)* symptoms is emerging as a problem in a sizeable subset of siblings of children with cancer (Alderfer & Hodges, 2010). Studies examining PTS reactions in siblings report that 29% to 38% exhibit moderate to severe posttraumatic stress, even years after cancer treatment has ended (Alderfer, Labay, & Kazak, 2003; Packman, 2004; Packman, Chesterman, vanZutphen, Golan, & Amylon, 2004; Packman, Crittenden, Schaeffer, et al., 1997).

Studies investigating the effects of pediatric cancer on siblings' *quality of life* have emerged in the last decade, revealing sample means indicative of poorer quality of life in siblings compared to norms (Houtzager et al., 2004; Houtzager, Grootenhuis, Hoekstra-Weebers, Caron, & Last, 2003; Houtzager, Grootenhuis, Hoekstra-Weebers, & Last, 2005; Packman et al., 2004; Packman et al., 2005).

In qualitative studies addressing siblings' *school functioning* (Freeman, O'Dell, & Meola, 2000; McGrath, Paton, & Huff, 2005; Nolbris, Enskar, & Hellstrom, 2007; Packman, Crittenden, & Rieger Fischer, 1997; Packman, Crittenden, Schaeffer, et al., 1997), disruptions in school performance, as well as a need to be more independent and responsible regarding homework were important issues (Alderfer et al., 2010). However, school performance also has been a topic in quantitative studies (e.g. (Dolgin et al., 1997; Houtzager, Grootenhuis, Hoekstra-Weebers, et al., 2005; Labay & Walco, 2004; Lahteenmaki, Sjoblom, Korhonen, & Salmi, 2004), showing that school problems among siblings might typically occur in the period of time that immediately follows diagnosis (Alderfer et al., 2010).

While several qualitative studies have identified increased *somatic complaints* in cancer siblings (Freeman et al., 2000; Grinyer & Thomas, 2001; Scott-Findlay & Chalmers, 2001; Sloper, 2000), quantitative findings investigating siblings somatic complaints or physical quality of life have yielded mixed results (Cuttini et al., 2003; Dolgin et al., 1997; Heffernan & Zanelli, 1997; Houtzager, Grootenhuis, Caron, & Last, 2005; Packman et al., 2004). In summary, somatic complaints and physical functioning do not seem to be poorer for most siblings, but very young siblings may be at some risk in the immediate aftermath of the cancer diagnosis (Alderfer et al., 2010).
Several studies have examined *resilience and positive outcomes* in cancer siblings (Heffernan & Zanelli, 1997; Packman, Crittenden, Schaeffer, et al., 1997; Phuphaibul & Muensa, 1999; Sloper, 2000; Wiener et al., 2008). Issues that have been revealed by qualitative and quantitative studies are increased responsibility, independence and maturity, as well as increased empathy, sensitivity, and compassion in siblings. However, according to Alderfer et al. (2010), more targeted research on these issues is required to understand the potential positive outcomes of the cancer experience.

4.2 Predictors of siblings adjustment

Several sibling, environmental and medical variables have been found to influence sibling adjustments to pediatric cancer. According to some (Houtzager et al., 2003; Sahler et al., 1994), a major risk factor for sibling adjustment problems is the presence of major life events prior to diagnosis. Further, a lack of social support seems to lead to poorer functioning in siblings (Barrera, Fleming, & Khan, 2004; Williams et al., 2002). Only a few studies have examined how time since diagnosis is relevant to adjustment. Preliminary results of review data show that the adjustment of siblings is more likely to be poorer than that of norms or controls closer to the time of cancer diagnosis (Alderfer et al., 2010). Considering age and gender, adolescents seem to exhibit the poorest adjustment, relative to other developmental stages, and females may exhibit more distress than males (Alderfer et al., 2010).

5. Intervention study at the Children's University Hospital Zurich

It has only been over the last few decades that the effects of childhood cancer on healthy siblings have begun to be explored. Nonetheless, researchers and clinicians in this field agree that siblings of children with cancer are a group at-risk for psychosocial problems. Hence, there is considerable interest in the development of early psychological interventions to support this vulnerable population. To design, implement and evaluate such an intervention was the main goal of the present PhD project. This venture is based upon a study on siblings of pediatric cancer patients that was conducted at Children's University Hospital in Zurich between December 2005 and December 2010. The author of the present doctoral thesis was

employed as a research fellow to conduct the study under the supervision of PD Dr. Markus A. Landolt and made crucial contributions to the study design. Furthermore, she was responsible for the development of the sibling interview and the questionnaires for parents, for the recruitment of families, for the acquisition of data, for statistical analyses, and for the composition of three journal articles that mainly constitute the present doctoral thesis. A UBS donation by client Ref. 14102004 financially supported the study at Children's University Hospital Zurich.

5.1 Objectives, research question, and hypothesis

The primary goals of the main project were to conceive, implement and evaluate a standardized, early, individual and two-session psychological intervention for school-aged siblings of pediatric cancer patients. We aimed at assessing the effect of our intervention by means of a randomized controlled trial. Our underlying concept was that early intervention might have the potential to prevent siblings from maladjustment subsequent to the cancer diagnosis of their brother or sister, quantifiable in terms of fewer symptoms of anxiety, depression and posttraumatic stress, by less problem behavior, by better health-related quality of life (HRQoL), and by enhanced social support.

Thus, we designed a study to yield evidence to answer the following research question and to support or invalidate the following hypothesis:

- Research question: Does our intervention improve siblings' psychological and social adjustment compared to a control group of siblings not receiving this intervention?
- Research hypothesis: Our approach will be effective in the sense of yielding evidence that siblings of cancer patients who receive the intervention suffer from fewer psychopathological symptoms, report a better HRQoL, and are better socially supported than a control group lacking this intervention.

Although a child experiencing a significant negative life event such as cancer in the family has a higher probability of developing psychological problems, the diagnosis alone is neither necessary nor sufficient to explain present or to predict future maladjustment among siblings (Cummings, Davies, & Campbell, 2000). Thus, we assume that the interplay of disease factors, family variables and sibling characteristics influence siblings' adjustment outcomes over time (see Figure 2). Accordingly, our intervention was intended to influence sibling and environmental predictor variables, so as to obtain a better outcome in siblings. Hence, it was designed to improve coping strategies, knowledge about illness and illness appraisal in siblings, and to install good social support for siblings in their environment (see variables in italics in Figure 2).

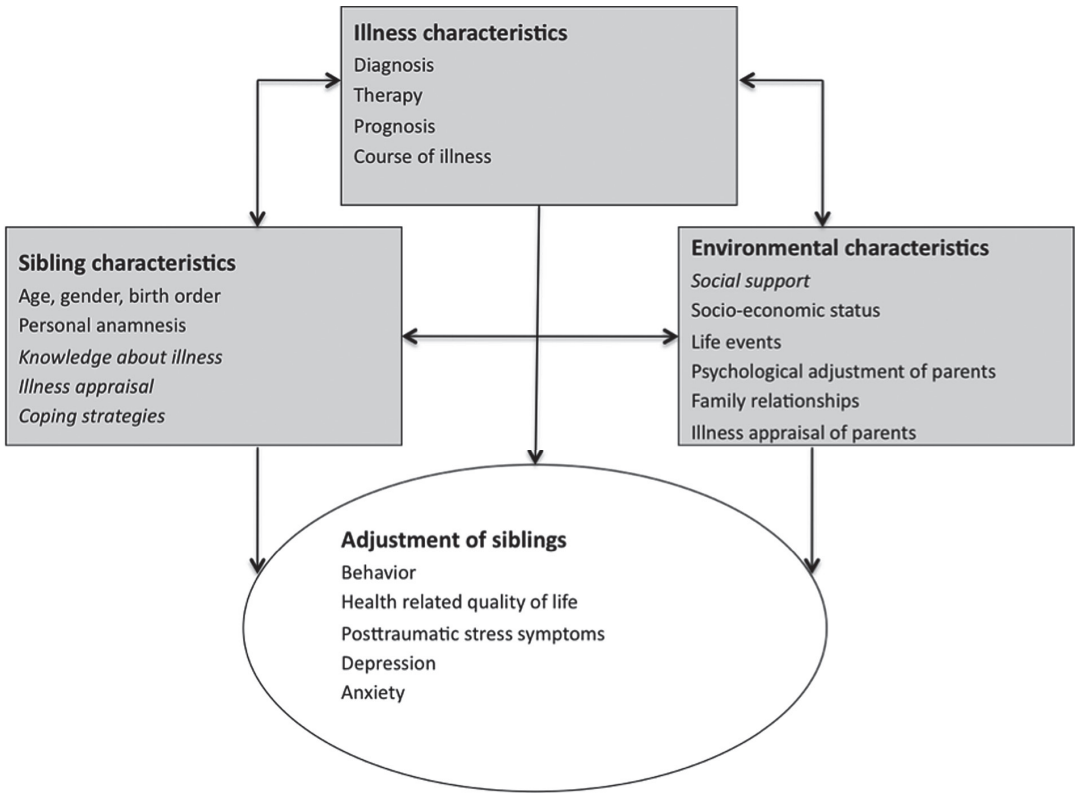

Figure 2. *Schematic diagram of the predictors of psychological adjustment in siblings (adapted from Landolt, 2003)*

5.2 Methods

5.2.1 The sibling intervention

The standardized psychological intervention was developed on the basis of clinical experience, theoretical considerations, the relevant literature, and a qualitative pilot study in which qualitative information about siblings' experiences over the first six months post-diagnosis was gathered (see Chapter III). It was designed as a individual intervention and took place within the first two months after the cancer diagnosis in the family. In two sessions, each approximately 50 minutes long, the author guided siblings and their parents through a three-step program.

First, siblings were given *medical information,* which is knowledge about body functioning, mechanism and location of the illness, and the cancer treatment, in particular chemotherapy. Second, emphasis was placed on *coping strategies.* Siblings were asked about their personal cancer-related stressors; and they learned how other siblings had appraised the stressors and were encouraged to develop their own coping strategies in response to their specific situation. Third, parents received a *psycho-educational booklet* that had been developed by the authors and contained information on the psychosocial situation of siblings of cancer patients in general, also providing recommendations to parents on how to support their healthy children (see Appendix A). A more detailed description of the intervention is provided in Chapter IV.

5.2.2 Procedures

To collect data for the present study, we used a randomized controlled trial (RCT), a study design that currently is being applied commonly in all areas of psychological and medical intervention research (Stinson, McGrath, & Yamada, 2003), leads to evidence-based results, and may provide clinicians valuable information about how to select the best treatment for individuals (Stinson et al., 2003). In the current study, participating siblings were randomly assigned to an intervention or control group. The manualized intervention was provided to siblings in the intervention group within the first 2 months after the cancer diagnosis. At 4 to 6 weeks (T0), 4 months (T1), and 7 months (T2) after the diagnosis, all siblings (in either the intervention or control group) were assessed by means of standardized face-to-face interviews and their parents completed questionnaires at home. The study procedure is described in Figure 3. Detailed information about participants and statistical procedures is given in Chapter IV.

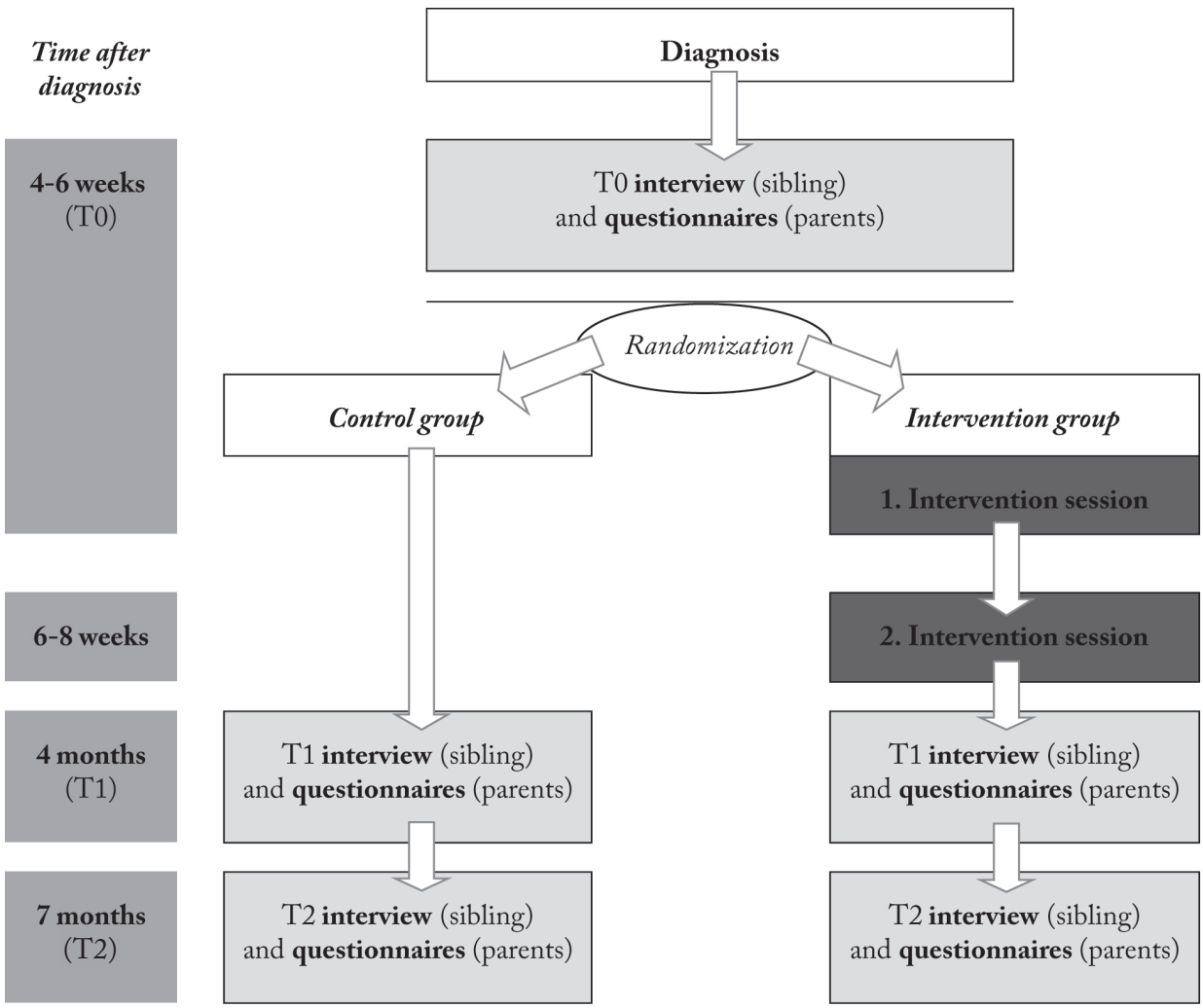

Figure 3. *Flowchart of the study design and procedure*

5.2.3 Measures

Information on siblings' adjustment, illness and environmental characteristics were collected by semi-structured interviews with siblings and questionnaires handed out to mothers, fathers and pediatricians in charge. Both the sibling interview and the questionnaires were comprised of standardized measures, as well as self-developed questions and scales.

The *sibling interview* assessed nine domains: 1) perception of the illness experience; 2) medical knowledge; 3) social support (satisfaction and amount); 4) health-related quality of life; 5) illness appraisal; 6) posttraumatic stress symptoms; 7) symptoms of depression; 8) symptoms of anxiety; and 9) coping strategies.

The *mothers' questionnaire* consisted of nine domains concerning the healthy sibling, as well as the mother herself and the family: 1) anamnestic information about the sibling (school, health, therapies); 2) behavioral problems in the sibling; 3) health-related quality of life in the sibling; 4) socio-economic status of the family; 5) psychiatric symptoms in the mother; 6) illness appraisal of the mother; 7) posttraumatic symptoms in the mother; 8) life events in the family; and 9) quality of the family environment.

The *fathers' questionnaire* consisted of five domains concerning the healthy sibling, as well as the father himself: 1) behavioral problems in the sibling; 2) psychiatric symptoms in the father; 3) illness appraisal of the father; 4) posttraumatic symptoms in the father; and 5) quality of the family environment.

The *questionnaire for the pediatrician* in charge consisted of questions regarding the cancer diagnosis, the intensity and kind of treatment, the length of hospitalization, medical complications, health-related restrictions, compliance of the patient, and information about relapse.

The measures used in the sibling interview and the parents' questionnaires are shown in Table 1. Detailed descriptions of those measures that were used in final data analysis are given in Chapter IV. Those measures that did not contribute to the final data analysis are described briefly in the present chapter, below.

Perception of impact of illness: The Sibling Perception Questionnaire (SPQ) by (Carpenter & Sahler, 1991) is a widely used measure for siblings' responses to childhood cancer. The SPQ measures school-aged siblings' perceptions of the illness' impact on a five-point Likert scale and contains four subscales: intrapersonal responses, interpersonal relationships, fear of disease, and communication about the illness. Taking the mean of all 23 items, a total problem score can be calculated. There are no normative data available for this measure, as of yet.

Illness appraisal: Subjective illness appraisal was assessed by three single items (appraisal of distress, threat and hope). These three items were derived from an appraisal scale that is comprised of seven different aspects of appraisal. The scale had been validated previously in pediatric patients (Vollrath, Landolt, & Ribi, 2004). Siblings and parents were asked about their subjective cancer-related distress level, about how serious and dangerous they appraise the ill child's illness, and how hopeful they tend to feel thinking about the ill child's future. The answer format was a three-point Likert scale (1–3) with different verbal descriptors for each level.

Depression: Depressive symptoms in siblings were assessed using the German version (Stiensmeier-Pelster, Schürmann, & Duda, 2000) of the Children's Depression Inventory (DIKJ, (Kovacs, 1985), a 26-item, standardized, self-rated symptom scale of depression. Scores range from 0 to 52 based upon item ratings of 0–2, with higher scores indicating greater severity of depression. A cut-off of 18 points has been shown to identify children with clinically relevant depression (Stiensmeier-Pelster, Schürmann, & Duda, 2000).

Coping strategies: To assess siblings' coping strategies, we used the German version (Rathner & Zangerle, 1996) of the KIDCOPE (Spirito, Stark, & Williams, 1988), a brief coping checklist comprised of 15 items. The checklist covers the following ten strategies: problem-solving, distraction, social support, social withdrawal, cognitive restructuring, self-criticism, blaming others, emotional expression, wishful thinking, and resignation. Using a four-point Likert scale, 0–3, siblings were asked how often they used a particular coping strategy when confronted with cancer-related stressors.

Behavioral problems: The Strength and Difficulties Questionnaire (SDQ) by (Goodman, 1997) is a brief behavioral screening questionnaire with 25 items generating scores for conduct problems, hyperactivity, emotional symptoms, peer problems, and pro-social behavior. All but the last score are summed to generate a total difficulties score. In the present study, the parent version was used to assess sibling behavioral problems.

Psychiatric symptoms in parents: The presence of psychiatric symptoms in parents was assessed using the Symptom-Checklist-27 (SCL-27) by (Hardt, Egle, Kappis, Hessel, & Brahler, 2004). The SCL-27 is a 27-item screening instrument that contains six symptom subscales: depressive, dysthymic, vegetative, agoraphobic, sociophobic, and symptoms of mistrust. Parents were asked how often they had suffered from certain symptoms over the last seven days. Responses were ranked on a five-point Likert scale from 0 (not at all) to 4 (most intensive).

Posttraumatic stress symptoms of parents: Posttraumatic stress reactions of parents were assessed using the Posttraumatic Diagnostic Scale (PDS, (Foa, 1997). This self-report measure of PTSD provides both a diagnosis according to DSM-IV criteria and a measure of PTSD symptom severity. It asks about 17 symptoms of PTSD that are rated on a 4-point Likert scale ranging from not at all (0) to very much (3). The questionnaire also includes one item that assesses the duration of the symptoms. In addition, nine items assess whether the reaction to the trauma has caused impaired functioning in different domains of life (yes/no format).

Life events in the family: We assessed the occurrence of 12 major life events in the family during the 12 months just prior to the cancer diagnosis and the six months following the diagnosis, as reported by the mother. This life event scale was developed at Children's University Hospital Zurich (Landolt & Vollrath, 1998) and has used in several other studies (Landolt, Vollrath, Gnehm, & Sennhauser, 2009; Landolt, Vollrath, & Ribi, 2002). The following life events were assessed: maternal pregnancy or a birth, parental divorce or separation, parental marriage, new person living in the household, significant change in parental income, serious indebtedness, change of domicile, change of job, unemployment, serious illness or accident

involving a family member (excluding the child with cancer), bereavement in the family or close friends, and change in a child's school.

Quality of family environment: Quality of family relationships was measured via the Family Relationship Inventory (FRI), which assesses the three relationship subscales cohesion, conflict, and expressiveness (Moos, 1994). Each scale is composed of nine items that traditionally are scored using a true-false format (0 or 1). For the purposes of our study, we extended the answer scale to a 3-point Likert scale consisting of the response options not true (0), partially true (1), and true (2).

Table 1. *Measures of the study*

Data source	Instruments
Sibling interview	• Perception of the illness experience: • Sibling Perception Questionnaire (SPQ) (Carpenter & Sahler, 1991) • Medical knowledge (self developed scale) • Social support (self developed scale) • Health related quality of life • KIDScreen child version (Ravens-Sieberer et al., 2005) • Illness appraisal (stress, threat, hope) (Vollrath et al., 2004) • Posttraumatic stress symptoms: • UCLA PTSD Reaction Index (UCLA RI) (Steinberg, Brymer, Decker, & Pynoos, 2004), German translation by University of Konstanz • Depression: • Children's Depression Inventory (DIKJ) (Stiensmeier-Pelster, Schürmann, M., Duda, K., 1989) • Anxiety: • Spence Children's Anxiety Scale (SCAS) (Spence, 1998) • Coping strategies: • KIDCOPE (Spirito et al., 1988); Germen version by (Rathner & Zangerle, 1996)
Questionnaire parents	• Anamnestic information about sibling * • Behavioral problems sibling: • Strengths and Difficulties Questionnaire (SDQ) (Goodman, Meltzer & Bailey, 2003) • Health related quality of life: * • KIDScreen Elternversion (Ravens-Sieberer et al., 2005) • Socio-economic status of family * (developed by the University Children's Hospital Zurich) • Psychiatric symptoms of parents: • Symptom-Checklist-27 (SCL-27) (Hardt et al., 2004) • Illness appraisal of parents (Vollrath et al., 2004) • Posttraumatic stress symptoms of parents: • Posttraumatic Diagnostic Scale (PDS) (Foa, 1997) • Life events in the family * (Landolt & Vollrath, 1998) • Quality of family environment: • Family Relationship Index (FRI) (Moos, 1994)

Questionnaire pediatrician	• Medical information: Diagnosis, intensity and kind of treatment, length of hospitalization, medical complications, health-related restrictions, compliance of patient and information about relapse (self developed questions)

** Only in the mother questionnaire*

6. Outline of this thesis

The present doctoral thesis is comprised of three papers, which all have been submitted to peer-reviewed journals for publication.

In Chapter II, a review of existing reports of psychological interventions with siblings of cancer patients is presented.

- Prchal, A. & Landolt, M. A. (2009). psychological interventions with siblings of pediatric cancer patients: a systematic review. *Psycho-Oncology,* 18, 1241–1251.

Chapter III describes the results of a qualitative study on how siblings of pediatric cancer patients experience the first six months after the diagnosis. Emphasis is on experiences in the hospital, in school, within the family, with peers, and with the ill child.

- Prchal, A. & Landolt, M. A. (in press). How siblings of pediatric cancer patients experience the first time after diagnosis: a qualitative study. *Cancer Nursing*

In Chapter IV, the results of an evaluation of our own intervention are presented.

- Prchal, A., Graf, A., Bergsträsser E. & Landolt, A. (under review). A two-session psychological intervention for siblings of pediatric cancer patients: a randomized controlled trial.

Finally, in Chapter V, all results are summarized and critically discussed, and clinical implications as well as suggestions for future research are given.

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II. Psychological interventions with siblings of pediatric cancer patients: a systematic review

1. Abstract

Objective: Siblings of pediatric cancer patients have been shown to be at risk for developing emotional, behavioral, and social problems. There is a need for psychological interventions in this population. Several researchers have previously documented and evaluated their interventions with siblings. This paper aimed at reviewing the existing reports of evaluated psychological interventions with siblings of pediatric cancer patients and at outlining future directions.
Methods: Research was conducted on several online bibliographic databases. Articles were selected on the basis of predefined criteria. If possible, effect sizes were calculated.
Results: Fourteen studies representing eleven different sibling interventions met criteria for inclusion. One individual intervention, three camps, and seven groups were found. Objectives of interventions concentrated mainly on enhancing siblings' coping and improving their medical knowledge. In terms of outcome measures, most of the studies focused on psychological adjustment variables. Findings showed significant improvements in siblings' depression symptomatology, medical knowledge, and health-related quality of life. Findings were inconsistent with regard to anxiety, behavioral problems, social adjustment, self-esteem, and posttraumatic stress symptoms. Depending on the outcome variables, small to large effect sizes were found. Satisfaction with the intervention was high in both siblings and parents.
Conclusion: There is tentative evidence that psychological interventions with siblings of childhood cancer patients can effectively reduce psychological maladjustment and improve medical knowledge about cancer. However, the number of studies is small, and several methodological shortcomings have to be noted. In future, more randomized controlled trials need to be conducted in larger samples to extend the evidence base.

2. Introduction

Diagnosis and treatment of childhood cancer is a stressful experience for all family members, including healthy siblings. As a result of the need to care for the ill child the entire family is forced to change daily life and routines. Understandably, the parents' attention becomes focused on their ill child, whereas healthy siblings are confronted with decreased availability of their parents (Spinetta, 1999). Siblings may also be worried about the illness, and they

have to observe their brother or sister undergo emotional and physical pain. Many siblings experience intrusive and conflicting emotions such as feelings of fear, isolation, jealousy, or guilt (Houtzager, Grootenhuis, Caron, & Last, 2004).

Most psychological research in pediatric oncology includes either patients alone or patients and mothers, with a lack of attention to siblings, fathers, or others in the child's social ecology (Kazak, 2005). This focus of research and literature on the sick child may reflect the siblings' position in the family during the illness process (Houtzager, Grootenhuis, & Last, 1999). However, the research on the psychological adjustment of siblings of cancer patients has grown over the past three decades. Houtzager et al. (1999) reviewed the literature on siblings' adjustment to childhood cancer and found that most siblings do not seem to suffer from severe psychopathology. But there is growing evidence indicating that siblings of children with cancer suffer significant psychological distress. *Internalizing problems* such as anxiety and depression (Bendor, 1990; Houtzager et al., 2004; Lahteenmaki, Sjoblom, Korhonen, & Salmi, 2004) as well as *externalizing behavioral problems* (Heffernan & Zanelli, 1997; Lahteenmaki et al., 2004) have been reported. Also, siblings suffer from *problems at school* (Carpenter & Sahler, 1991; Labay & Walco, 2004), *psychosomatic complaints* (Heffernan & Zanelli, 1997; Lahteenmaki et al., 2004; Zeltzer, 1996), lower *quality of life* (Houtzager, Grootenhuis, Hoekstra-Weebers, Caron, & Last, 2003; Lahteenmaki et al., 2004), and *post-traumatic stress* (Alderfer, Labay, & Kazak, 2003; Packman, 1999). Yet, siblings have also reported some *positive effects,* including maturation, understanding, compassion, and closer family relationships (Sargent et al., 1995; Sloper, 2000).

In sum, siblings of pediatric cancer patients have been shown to be at risk for developing several emotional, behavioral, and social problems, and most clinicians and researchers agree that there is a need for psychological interventions for this population (Alderfer & Noll, 2006; Chung, Miranda, Fleming, & Barrera, 2004). However, the majority of published papers on siblings of pediatric cancer patients still report the results of descriptive and correlational research, and there is a lot of non-empirical, anecdotal data in this field. Similar to the whole field of pediatric psycho-oncology, intervention research in siblings is rare (Kazak, 2002). Nevertheless, several authors have documented sibling programs and have examined the effects of the interventions. The present paper reviews empirical evidence on the effectiveness of psychological interventions with siblings by summarizing outcome variables and calculating effect sizes (ES) whenever possible. Further, the review describes objectives and settings of existing interventions and gives recommendations for future research in this field.

3. Method

3.1 Data sources and search strategy

Relevant electronic databases were searched for published empirical studies and dissertations evaluating psychological interventions with siblings of pediatric cancer patients. The databases searched were Medline, EMBASE, CINAHL, PsycINFO, Cochrane Systematic

Reviews, and ProQuest Digital Dissertation. Searches were conducted in September 2008 for the period 1980–2008.

The searches were carried out using the following search terms for the patient population: (1) *sibling* (in title); (2) *pediatric, paediatric, child, adolescent;* (3) *neoplasm, oncology, cancer.* Search terms for the intervention were *intervention, camp, group, weekend, support, counsel, education, therapy* (in title or abstract). The Boolean operator "and" was used to combine the three patient population identifiers and identifiers for intervention. The operator "or" was used to combine identifiers within the three population areas and the intervention area.

The initial literature search yielded 449 hits with 431 articles and 18 dissertations. In a preselection process, completed by the first author, titles and abstracts were screened for existence of psychological interventions. Thirty-five articles and dissertations remained. First and second author checked the full text of pre-selected studies for inclusion criteria according to a standardized checklist and obtained agreement in case of uncertainty. Additionally, reference lists of relevant studies and reviews were examined to identify other pertinent articles. Doctoral dissertations were examined only if they were accessible in full text.

3.2 Study selection

Under examination were studies evaluating the effect of interventions aimed at improving the psychological adjustment of siblings of childhood cancer patients. Studies were included if they met the following criteria: (a) *type of studies:* evaluation of an intervention with quantifiable data about the effect of the intervention, (b) *type of outcome measures:* standardized and validated outcome measures with self or proxy report of psychological adjustment and/or quality of life and/or satisfaction with intervention and/or medical knowledge, (c) *types of participants:* children and adolescents age 0 to 18 years, sister or brother diagnosed with childhood cancer, not more than 50% bereaved children in the sample, sample size at least 10, and (d) *type of intervention:* standardized and sibling specific intervention program, content of intervention described, specific for childhood cancer. Sibling programs with a majority of bereaved children were excluded because we assumed that they focus mainly on grief and loss rather than on coping with specific sibling stressors. If necessary, corresponding authors were contacted for additional information or clarification of the text. Reasons for exclusion were recorded (Table 2). Most papers (62%) had to be excluded because outcome measures were not standardized and validated.

3.3 Data extraction

Data was extracted by the main author from each publication into a standardized data collection form. Information about study design, sample size, age and months since diagnosis were recorded. Further, information about the specific sibling interventions (setting, number of sessions) and about outcome variables were extracted. We subdivided outcome variables into variables assessing psychological adjustment (depression, anxiety, social adjustment, self-esteem, behavioral problems, and posttraumatic stress symptoms) and quality of life. In ad-

dition, we examined findings based on the Sibling Perception Questionnaire (SPQ) by Carpenter and Sahler (1991), a widely used measure for siblings' responses to childhood cancer. The SPQ was developed to assess school-aged siblings' responses in the three domains *perceptions of impact of the illness* (subscales intrapersonal responses, interpersonal relationships, fear of disease, and communication about the illness), *affective responses* (positive and negative mood), and *medical knowledge* (instrumental and identification).

Table 2. *Excluded studies*

Study	Reason for exclusion
Adams-Greenly et al., 1986	1
Ballard, 2004	1; 2
Bedway & Hartkopf Smith, 1996	1; 2; 3
Bordeur, 2005	1; 2; 4; intervention unclear
Bendor, 1986	1;2; age group up to 22y
Carpenter, Sahler & Davis, 1990	Same intervention, data set and outcome variables as Sahler & Carpenter, 1989 [26]
Creed, Ruffin, & Ward, 2001	1; only bereaved children
Cunningham, Betsa, & Gross, 1981	1
Fanos et al., 2005	1; 3 (percentage of childhood cancer patients unclear); age unclear
Kazak, 1999	Sample size too small
Kinrade, 1985	1
Kramer & Moore, 1983	1
Lobato & Kao, 2002	3
Lobato & Kao, 2005	3 (cancer population 14 %)
Olin, 1998	Outcome measure: stress measured by cortisol concentration in children's saliva, unclear if sibling-specific intervention
Packman, 2008	1
Ruffin, Creed, & Jarvis, 1997	1;4
Simms et al., 2002	1;2
Wellisch et al., 2006	Unclear if sibling-specific part existent
Williams et al., 1997	3 (cancer population 36.4 %)
Williams et al., 2003	3 (cancer population 8.7 %)

1 Outcome measure not standardized / validated
2 Intervention not standardized
3 Intervention not specific for childhood cancer
4 Intervention not sibling specific

There are no normative data available for this measure as yet, but the instrument is being used increasingly in studies with siblings of children with cancer (Sidhu, Passmore, & Baker, 2006). Finally, siblings' and parents' satisfaction with the intervention program was an outcome variable examined (all self-developed scales).

The comparison of ES across different studies makes it possible to judge the strength of the finding across different interventions, sample characteristics, or methods. Unfortunately, the data to be analyzed for computing ES are often not readily available from the study reports. Of the reviewed studies only two (Barrera, Chung, & Fleming, 2004; Packman, Chesterman, vanZutphen, Golan, & Amylon, 2004) reported ES in their original papers. Pre/post ES (Cohen's d) were calculated as the within-group difference between pre-intervention and post-intervention means divided by the weighted pooled pre/post standard deviation for the intervention group. Also, between-group ES were computed whenever possible, comparing the difference between pre/post-mean for the experimental and the comparison group divided by the pooled standard deviation. All ES were calculated by the present author from the data given in the research reports, whenever the necessary information was available. Positive ES represent improvements in the desired direction. Cohen (1992) suggests that an ES of 0.2 is indicative of a small, 0.5 of a medium, and 0.8 of a large effect.

4. Results

Fourteen studies (ten published articles and four dissertations) met the inclusion criteria, representing eleven different sibling interventions. Two studies (Packman et al., 2004; Packman et al., 2005) are based on the same dataset but present other outcome variables. The main characteristics of the included studies are summarized in Table 3. ES are presented in Table 4.

4.1 Description of studies

Origin: The studies reviewed were published between 1986 and 2005. Eight publications are from the United States (Atherton, 1984; Barrera et al., 2004; Barrera, Chung, Greenberg, & Fleming, 2002; Chung, 2000; Cimini, 1986; Dennis, 1995; Heiney, Goon-Johnson, Ettinger, & Ettinger, 1990; Kazak et al., 2004; Packman et al., 2004; Packman et al., 2005; Sahler & Carpenter, 1989), three from Canada (Barrera et al., 2004; Barrera et al., 2002; Chung, 2000), and one each from Israel (Dolgin, Somer, Zaidel, & Zaizov, 1997), the Netherlands (Houtzager, Grootenhuis, & Last, 2001), and Australia (Sidhu et al., 2006), respectively.

Study design: Ten studies used pre-post intervention designs. Only three publications are based on randomized, controlled trials (RCTs) to evaluate the intervention (Atherton, 1984; Cimini, 1986; Kazak et al., 2004). One study provided a non-randomized control group in addition to the pre- and post evaluation (Heiney et al., 1990).

Sample characteristics: The sample sizes varied from 11 to 90 siblings with an age range from 6 to 20 years. All studies but one assessed the effect of the intervention exclusively among siblings. Kazak et al. (2004) evaluated an intervention with cancer survivors and their fami-

lies including siblings, but they used sibling-specific material when working with siblings. Time since onset of cancer in the ill child varied within and between all studies from months to several years. Five publications did not report time since diagnosis (Barrera et al., 2004; Barrera et al., 2002; Heiney et al., 1990; Packman et al., 2004; Packman et al., 2005).

Table 3. *Summary of studies included in the review*

Study	Design	N	Age (years)	Months since diagnosis	Setting	Sessions	Intervention objective	Outcome measures
Atherton, 1984	rct	18 (9/9)	8–13	<6	single	2 (30 min)	medical knowledge coping	= anxiety (s) [b]
Barrera et al., 2002 [1]	pp	12	6–17	–	group	8 (120 min)	medical knowledge coping	+ depression (s) [a] + anxiety (s) / = anxiety (p) [b] = behavior (s, p) [k j] + SPQ (s) / = SPQ (p) [o] + satisfaction (s, p) [q]
Barrera et al., 2004 [1]	pp	47	6–14	–	group	8 (120 min)	medical knowledge coping	+ depression (s) [a] + anxiety (s, p) [b] = behavior (s) / + behavior (p) [k j] + satisfaction (s, p) [q]
Chung, 2000 [1]	pp	25	6–17	1–128	group	8 (120 min)	medical knowledge coping	+ depression (s) [a] = anxiety (s) / + anxiety (p) [b] = self-esteem (s) [g] = behavior (p) [j] + SPQ (s) / -SPQ (p) [o] + satisfaction (s, p) [q]
Cimini, 1986	pp; rct	30 (15/15)	7–14	<12	group	3 (1 day)	medical knowledge coping	+ depression (s) [a] + anxiety (s) [c d] = behavior (p) [j] + satisfaction (s) [p q]
Dennis, 1995	pp	11	8–13	3–36	group	10	medical knowledge coping family communication	= anxiety (s, p) [c] = self-esteem (s) [h] + SPQ (s) [o] + satisfaction (s) [p q]

Table 3. *Summary of studies included in the review (continued)*

Study	Design	N	Age (years)	Months since diagnosis	Setting	Sessions	Intervention objective	Outcome measures
Dolgin et al., 1997	pp	24	7–17	4–48	group	6	medical knowledge coping family communication	+ SPQ (s) [o] + satisfaction (s) [q]
Heiney et al., 1990	pp; ct	14	9–15	–	group	7	medical knowledge coping	= social adjustment (s) [f] + satisfaction (s) [q]
Houtzager et al., 2001	pp	24	7–18	2–89	group	5	medical knowledge coping	+ anxiety (s) [b]
Kazak et al., 2004	rct	43 (19/24)	10–20	12–146	group	4 (1day)	coping family communication PTS reduction	= anxiety (s) [c] = PTS (s) [l]
Packman et al., 2004 [2]	pp	77	6–17	–	camp	1 week	coping recreational	+ anxiety (s) [c] + self-esteem (s) [l] + PTS (s) [m] + HRQoL (s) [n]
Packman et al., 2005 [2]	pp	77	6–17	–	camp	1 week	coping recreational	+ HRQoL (s) / = HRQoL (p) [n]
Sahler & Carpenter, 1989	pp	90	6–17	4–156	camp	5 days	medical knowledge coping recreational	+ SPQ (s) [o]
Sidhu et al., 2006	pp	26	8–13	app. 12	camp	4 days	medical knowledge coping recreational	+ anxiety (s) [e] + social adjustment (s) [g] +self-esteem (s) [g] + SPQ (s) [o]

[1] = same intervention and but different data set; [2] = same intervention and data set but different outcome variables , rct = randomized controlled trial, ct = controlled trial; pp = pre and post intervention assessment; s = self report; p = proxy report , + indicates significant improvement in the desired direction compared to control / pre assessment; - indicates significant impairment compared to control / pre assessment, = indicates no significant difference between intervention and control group or no significant change from pre to post assessment

[a] Children's Depression Inventory (CDI); [b] State-Trait Anxiety Inventory for Children (STAIC); [c] Revised Children's Manifest Anxiety Scale (RCMAS); [d] Fear Survey Schedule for Children Revised (FSSC-R); [e] Self-Report of Personality (SRP), Subtest of Behavior Assessment System for Children (BASC); [f] Social Adjustment Scale (SAS); [g] Self-Perception Profile for Children (SPPC); [h] Piers-Harris Children's Self-Concept Scale; [i] Rosenberg Self-Esteem Scale (RSE); [j] Child Behavior Checklist; [k] Youth Self-Report (YSR); [l] Post-Traumatic Stress Disorder Reaction Index (PTSD-RI); [m] UCLA PTSD Index; [n] Pediatric Quality of Life Inventory (PedsQL); [o] Sibling Perception Questionnaire (SPQ); [p] No psychometric evaluation; [q] Self-developed scales

Table 4. *Effect sizes d and mean effect sizes*

	Depression	Anxiety		Behavior		PTS	HRQoL		SPQ intrapersonal		SPQ interpersonal		SPQ communication		SPQ fear of disease	
	s	*s*	*p*	*s*	*p*	*s*	*s*	*p*	*s*	*p*	*s*	*p*	*s*	*p*	*s*	*p*
Barrera et al., 2002	0.47	0.98	0.68	1.45	0.35				1.12	-0.11	0.62	0.05	0.31	1.45	1.0	-0.19
Barrera et al., 2004	0.40	0.48	0.69													
Chung, 2000	0.31	0.36	0.95		0.39				0.65	0.19	0.11	0.11	0.22	0.27	0.48	-0.48
Dolgin et al., 1997									1.37		0.87		0.57		0.0	
Kazak et al., 2004		0.50														
Packman et al., 2004, 2005		0.44				0.47	0.32	0.12								
Mean effect size of outcome variable	0.40	0.55	0.77	1.45	0.37	0.47	0.32	0.12	1.05	0.04	0.53	0.08	0.37	0.86	0.49	-0.34

s = self report; p = parent report

PTS = posttraumatic stress; HRQoL = Heath-related Quality of Life; SPQ = Sibling Perception Questionnaire

4.2 Types of interventions

Setting: Seven of the 11 different intervention programs used a group setting (Barrera et al., 2004; Barrera et al., 2002; Chung, 2000; Cimini, 1986; Dennis, 1995; Dolgin et al., 1997; Heiney et al., 1990; Houtzager et al., 2001; Kazak et al., 2004), with three to ten sessions and group size from four to twelve siblings. Three interventions were provided in a camp format (Packman et al., 2004; Packman et al., 2005; Sahler & Carpenter, 1989; Sidhu et al., 2006) with camp duration of four to seven days. Only one intervention was designed as an individual intervention (Atherton, 1984). In two intervention programs parents were involved (Dennis, 1995; Kazak et al., 2004).

Lead Professional: The interventions were provided by a variety of professionals, including psychologists (Barrera et al., 2004; Barrera et al., 2002; Chung, 2000; Cimini, 1986; Houtzager et al., 2001; Packman et al., 2004; Packman et al., 2005), nurses (Atherton, 1984), or interdisciplinary teams (Dennis, 1995; Dolgin et al., 1997; Heiney et al., 1990; Kazak et al., 2004; Sahler & Carpenter, 1989; Sidhu et al., 2006).

Theoretical basis: Description of interventions in the selected publications varied considerably with respect to details, content, and procedure. Various theoretical frameworks were given as basis for the sibling intervention. Some authors designed their program on the basis of psychotherapeutic conceptual models such as CBT (Barrera et al., 2004; Barrera et al., 2002; Chung, 2000), group therapy (Dennis, 1995; Heiney et al., 1990), or family therapy (Dennis, 1995; Heiney et al., 1990). Others conducted a pre-intervention survey about sibling's needs (Dolgin et al., 1997; Sidhu et al., 2006) or chose their specific content areas based on clinical and research literature (Atherton, 1984; Cimini, 1986; Packman et al., 2004; Packman et al., 2005; Sahler & Carpenter, 1989).

Objectives: To examine specific types of intervention, we divided the reviewed studies into five categories according to the objectives of the program: Enhancement of (1) medical knowledge, (2) coping, and (3) family communication, (4) reduction of posttraumatic stress, and (5) providing peer support through the experience of recreational activities. Various types of intervention objectives were found. Enhancement of coping with sibling-specific stressors and emotions was an important objective in all selected intervention programs. Nine of the examined interventions focused also on improvement of medical knowledge (Atherton, 1984; Barrera et al., 2004; Barrera et al., 2002; Chung, 2000; Cimini, 1986; Dennis, 1995; Dolgin et al., 1997; Heiney et al., 1990; Houtzager et al., 2001; Sahler & Carpenter, 1989; Sidhu et al., 2006), that is, siblings were provided with information about pediatric cancer and its treatment. Three intervention programs tried to achieve better family communication (Dennis, 1995; Dolgin et al., 1997; Kazak et al., 2004); one program aimed to reduce posttraumatic stress (Kazak et al., 2004). Finally, three interventions, all in a camp setting, had their major focus on recreational activities (Packman et al., 2004; Packman et al., 2005; Sahler & Carpenter, 1989; Sidhu et al., 2006) in order to provide siblings with peer support.

Time since onset of cancer: Three programs defined time since onset of cancer in the ill child as an inclusion criteria for participating siblings. The intervention presented by Cimini (1986) required the patient to be under treatment for at least one year, Kazak et al. (2004) wanted

patients to have completed treatment for at least one year. Only the intervention evaluated by Sidhu et al. (2006) targeted siblings of children with cancer in an early stage of medical treatment and therefore has a preventive character.

4.3 Effectiveness of interventions

4.3.1 Psychological adjustment

Self-reported **depression** was assessed in four studies (Barrera et al., 2004; Barrera et al., 2002; Chung, 2000; Cimini, 1986), including one RCT study (Cimini, 1986), all of them using the Children's Depression Inventory (CDI) (Kovacs, 1992). The results suggest a significant reduction in siblings' depressive symptoms after the intervention. All the corresponding studies were provided in a group format, and three are based on the same intervention program (Barrera et al., 2004; Barrera et al., 2002; Chung, 2000). Effect sizes range from 0.31 to 0.47, indicating a small beneficial effect. No study assessed proxy-reported depression.
Ten publications used self-reported **anxiety** as an outcome measure. The majority of studies assessed anxiety using either the State-Trait Anxiety Inventory for Children (STAIC) (Spielberger, 1983) or the Revised Children's Manifest Anxiety Scale (RCMAS) (Reynolds & Richmond, 1985). Six publications (including one RCT) found a reduction in self-reported anxiety due to the intervention (Barrera et al., 2004; Barrera et al., 2002; Cimini, 1986; Houtzager et al., 2001; Packman et al., 2004; Sidhu et al., 2006); four studies (including two RCTs) did not find significant differences (Atherton, 1984; Barrera et al., 2002; Chung, 2000; Dennis, 1995; Kazak et al., 2004). No conclusive differences between studies with or without anxiety reduction could be found concerning intervention setting or number of subjects. Proxy-reported anxiety decreased in two studies (Barrera et al., 2004; Chung, 2000) and showed no significant differences in two other studies (Barrera et al., 2002; Dennis, 1995). All four studies were in group format and used a pre-post intervention design; however, the two with a decrease of anxiety included more subjects. ES for anxiety among all studies allowing calculation varied from small to large (ES 0.36–0.98).
Two articles evaluated self-reported **social adjustment** before and after the intervention. No significant improvement could be found by Heiney et al. (1990), using a non-equivalent control group, whereas the other study by Sidhu et al. (2006) reported improvement between pre and post measurements. No ES were available for measures of social adjustment.
Sibling-rated **self-esteem** was examined in four publications. Two using a camp format found enhanced self-esteem ratings after participation in a sibling intervention (Packman et al., 2004; Sidhu et al., 2006). Two studies in group format reported no significant change (Chung, 2000; Dennis, 1995). Sample sizes were larger in the studies with enhanced self-esteem. No ES were available for measures of self-esteem.
Self-reported and proxy-reported **behavioral problems** were assessed in four publications, all using the Child Behavior Checklist (CBCL) for parents or the Youth Self-Report (YSR) for adolescent siblings' reports (Achenbach, 1991). With the exception of Barrera et al. (2004), who found a significant reduction in parents' report of behavior problems from post-inter-

vention to follow-up, none of the other studies (Barrera et al., 2002; Chung, 2000; Cimini, 1986), including one RCT, found any significant changes in siblings' behavioral adjustment, and only trends in the desired direction were reported. Little data was available for calculating ES (ES 0.35–1.45). The very large ES of 1.45 for siblings' self-reported behavior assessed by Barrera et al. (2002) must be interpreted with caution, because this figure is based on a subset of only seven siblings.

Two of the reviewed studies used self-reported **posttraumatic stress** (PTS) as outcome variables (Kazak et al., 2004; Packman, 2004) assessed using either the Child Post-Traumatic Stress Disorder Reaction Index (Pynoos et al., 1987) or the UCLA PTSD Index (Rodriguez, Steinberg, & Pynoos, 1998). Findings were inconsistent. Whereas Packman et al. (2004) presented a significant reduction in posttraumatic stress symptoms from pre to post in their camp intervention, no changes could be found in the RCT by Kazak et al. (2004), who conducted a group intervention. ES could be computed only for one study (Packman et al., 2004) showing a small effect (ES 0.47).

Eleven of the 14 reports focused mainly on psychological adjustment variables as outcome measures. Proxy-raters were parents in all cases. Whereas the majority of studies report significant changes in at least one of the observed adjustment variables, four studies did not find changes in siblings' psychological adjustment after treatment (Atherton, 1984; Dennis, 1995; Heiney et al., 1990; Kazak et al., 2004).

4.3.2 Health-related quality of life

The influence of the intervention on siblings' quality of life was examined in two publications (Packman et al., 2004; Packman et al., 2005), both using the Pediatric Quality of Life Inventory (PedsQL) (Varni, Seid, & Rode, 1999), an instrument measuring **health-related quality of life** (HRQoL) in children and adolescents. Both studies showed significant improvements in self-reported HRQoL from pre- to post-camp. Packman et al. (2005) found significant changes in proxy reports from non-bereaved parents. Calculated ES (data from only one study available) were small (ES 0.32) for self reports and showed no effect for parents' reports.

4.3.3 Sibling Perception Questionnaire

The **Sibling Perception Questionnaire** (SPQ) (Carpenter & Sahler, 1991) was used in six publications, and all of them found significant changes in the desired direction in at least one of the SPQ subscales. Only two of the studies (Dolgin et al., 1997; Sahler & Carpenter, 1989) used all three domains of the SPQ, whereas others applied only the domain "perception of impact of illness" (Barrera et al., 2002; Chung, 2000; Sidhu et al., 2006) or the "medical knowledge" domain (Dennis, 1995).

Siblings' ***affective responses*** (positive and negative mood) improved significantly after the intervention in sibling and parent reports (Dolgin et al., 1997; Sahler & Carpenter, 1989). Results also indicated significant beneficial effects in ***medical knowledge*** as reported by siblings (Dennis, 1995; Dolgin et al., 1997; Sahler & Carpenter, 1989).

Regarding the domain ***perception of impact o the illness*** the findings are inconsistent. In the sibling-rated *intrapersonal* subscale, a measure of how illness affects the individual, significant improvements after intervention could be demonstrated in four (Barrera et al., 2002; Chung, 2000; Dolgin et al., 1997; Sahler & Carpenter, 1989) out of five studies. Parent reports showed no significant amelioration in intrapersonal responses to illness (Barrera et al., 2002; Chung, 2000). The available ES reinforced these results with medium to large ES for sibling-reported intrapersonal responses (ES 0.65−1.37) and with no effect in parent-reported intrapersonal responses. In the *interpersonal* subscale, a measure of interactions and relationships with others, three publications found no significant changes in sibling reports (Barrera et al., 2002; Chung, 2000; Sahler & Carpenter, 1989), and two found significant improvement (Dolgin et al., 1997; Sidhu et al., 2006). Parent reports again showed no improvement after the intervention (Barrera et al., 2002; Chung, 2000). ES for self-reported interpersonal relationships showed a wide range from no effect to large intervention effects (ES 0.11−0.87); based on parent reports, ES were negligible. Concerning the subscale *communication about illness* only one study found significant improvements in sibling reported communication (Dolgin et al., 1997); four other publications failed to find a significant change (Barrera et al., 2002; Chung, 2000; Sahler & Carpenter, 1989; Sidhu et al., 2006). Barrera et al. (2002) found significant improvements in parents' appraisal of communication, but Chung (2000) in turn could not find such improvements. ES for sibling and parental report of communication were small to large (ES 0.22−1.45). Finally, evaluation results concerning the SPQ subscale *fear of disease* in siblings were inconsistent among the studies. Remarkable is a significant increase in siblings' fear of disease reported by parents in the study by Chung (2000). According to this, ES varied considerably in self-reported fear of disease (ES 0−1) and showed no effect or a negative effect in proxy reports.

4.3.4 Satisfaction with intervention

Satisfaction measures are used to obtain the participants' feedback about the value and quality of an intervention program. Seven studies applied quantitative measures of satisfaction, all administered at the final session or at follow-up (Barrera et al., 2004; Barrera et al., 2002; Chung, 2000; Cimini, 1986; Dennis, 1995; Dolgin et al., 1997; Heiney et al., 1990). In four studies data was collected from both parents and siblings (Barrera et al., 2004; Barrera et al., 2002; Chung, 2000; Cimini, 1986). Overall, four different satisfaction instruments were used. Only the instrument used by Barrera et al. (2004; 2002) and Chung (2000) was tested with regard to its reliability and validity. Overall, satisfaction measures revealed in both siblings and parents a generally high level of satisfaction with the interventions provided. In addition to the quantitative evaluation three studies conducted a qualitative analysis of intervention effects on the basis of semi-structured interviews with siblings and parents (Chung, 2000; Packman et al., 2005) or follow-up focus groups with parents (Sidhu et al., 2006). On these qualitative measures both parents and siblings reported a clearly positive impact of the sibling intervention, thus underlining the results from quantitative satisfaction surveys. Findings from the qualitative evaluation suggested increased communication between siblings

and better medical knowledge about cancer in siblings as well as a decreased sense of isolation (Chung, 2000). Packman et al. (2005) found that the majority of responses emphasized the importance of group cohesiveness, shared personal experiences, socializing, and the relieving of emotions by expressing one's feelings.

4.3.5 Predictors of intervention effects

In seven studies predictors of intervention effects were examined (Barrera et al., 2004; Barrera et al., 2002; Cimini, 1986; Houtzager et al., 2001; Packman et al., 2004; Packman et al., 2005; Sahler & Carpenter, 1989). They discuss the influence of age, sex, diagnosis, bereavement, number of camp attendances, or pre-existing behavioral problems on intervention effects. Houtzager et al. (2001) found anxiety in siblings of children with leukemia or lymphoma decreased more after group participation than anxiety in siblings of children with a solid or a brain tumor. Barrera et al. (2004) identified age and gender as crucial factors influencing the effect of treatment, indicating that young boys are the most receptive to their intervention with greatest reduction in depression. In comparison, Cimini (1986) examined the impact of age and gender on measures of anxiety and depression and could not find any differences. Sahler & Carpenter (1989) found that with respect to medical knowledge, their camp seemed to be most beneficial to younger and returning campers. Further, they reported that siblings with behavioral problems seemed to benefit most from attendance at a camp. Packman et al. (2004) found that there was a slightly higher benefit for first time campers regarding posttraumatic stress and HRQoL compared to siblings who had participated in a camp previously. Finally, Packman et al. (2005) revealed that bereaved parents reported siblings' HRQoL as less positively influenced by a camp as compared to non-bereaved parents.

5. Discussion

5.1 Summary of results

This study aimed at reviewing existing reports on the effects of psychological interventions with siblings of pediatric cancer patients. Fourteen studies representing eleven different sibling interventions met criteria for inclusion. The reviewed studies included ten pre-post evaluations, three RCTs and one non-randomized control group design. Three interventions were conducted in camp settings and seven in group settings; one study described an individual intervention.

Looking at the intervention setting, it may be surprising that individual interventions are so rare, since sibling programs need to be developmentally appropriate and relevant in addressing personal concerns and individualized cancer-related information. These goals seem to be best met in an individual context in which one can emphasize on the needs of the sibling. On the other hand, the use of a group or camp format can give siblings the sense that they are not alone, and meeting others with similar experiences can itself have a positive impact on psychological adjustment. Further, the use of a group or camp format is cost effective.

Unfortunately, the existing studies do not allow a comparison of the efficacy of group versus individual format. The only single intervention included in the review showed no changes in psychological adjustment. In other fields of clinical child psychology, group and individual treatments could be shown to be equally effective in reducing children's affective symptoms (Hoag, 1997; In-Albon & Schneider, 2007).

Outcome evaluations revealed improvements in depressive symptomatology, negative mood, medical knowledge, and HRQoL in siblings following the intervention. Findings were inconsistent with regard to anxiety, behavioral problems, social adjustment, self-esteem, posttraumatic stress symptoms, and perception of the illness. Four out of 14 studies found no quantitative change in outcome variables after the intervention. These four studies represent different study designs, settings, and intervention objectives.

These findings need to be interpreted with caution because just a minority of studies used control groups to evaluate their outcome. However, by only looking at results of RCTs, findings are still inconsistent. Two out of three RCTs found no significant changes in their outcome variables (Atherton, 1984; Kazak et al., 2004); one RCT reported amelioration in depressive symptoms and anxiety post intervention (Cimini, 1986).

The reduction of depressive symptoms and amelioration of mood state in the present review leads to the assumption that sibling interventions have a positive influence on siblings' emotional condition. However, the ES for depressive symptoms were rather small. Based on the assumption that siblings that are provided with accurate medical information cope with and adapt to the situation more effectively (Evans, Stevens, Cushway, & Houghton, 1992; Havermans & Eiser, 1994), enhancement of medical knowledge was an important goal in the majority of the intervention programs reviewed. Only three studies assessed siblings' knowledge before and after the intervention, but they all found significant improvement on this variable. Increase in medical knowledge can also be found after interventions for siblings of children with a chronic illness or a disability (Lobato & Kao, 2002; Williams et al., 2003). There is tentative evidence that by gaining information about illness and treatment, siblings may experience enhanced feelings of control and therefore feel more secure and less anxious (Drotar & Crawford, 1985; Sourkes, 1980). In the present review anxiety was the outcome measure most often used, but only half of the studies found an anxiety reduction post intervention.

Depending on outcome variables different effect sizes were found. Out of 37 ES calculated, 28 showed small to large effects (ES $\geq$ .20) and 8 showed no effect of intervention. Only one negative effect was found (Chung, 2000). The issue of parent-reported significant increase in fear of cancer found by Chung (2000) might be a result of the siblings discussing their fears of cancer with their parents more openly after their participation in the sibling intervention. This explanation was supported by qualitative analyses conducted (Chung, 2000). In general, fewer proxy reports were given, and parents tend to estimate intervention effect less positively as compared to siblings.

Despite great diversity in their design, objectives, and lead professionals, sibling interventions produced positive effects on psychological adjustment, siblings' knowledge, and HRQoL.

With respect to siblings' and parents' satisfaction with the intervention, the bulk of evidence is quite positive. The overall results of the present review support the efficacy of sibling interventions and reveal no significant negative effects.

5.2 Limitations of previous studies

There are several methodological issues that limit generalization of the findings. First, ten out of fourteen publications evaluated intervention effects with a sample size smaller than 50. Several researchers complained that they could not recruit a larger number of participants due to the small number of potential subjects available in this population. Second, only a minority of studies used RCTs even though this is the gold standard for intervention evaluations (Chambless & Hollon, 1998; Spirito, 1999). A problem with the widely used pre-post design is the fact that positive changes of outcome measures may be due to natural effects of time or maturation rather than intervention effects. Several researchers explained the absence of a control group citing ethical and practical reasons. With regard to the ethical concerns of not providing siblings in a control group with the opportunity to participate in a intervention, there is still the possibility of a waiting list design or two assessments before providing the intervention. Third, few authors reported ES, and some did not even deliver relevant data in their report to calculate ES; thus, comparison across different types of interventions is difficult. Fourth, the great variety of outcome measures and instruments make comparisons quite difficult, and predictors have been assessed only unsystematically. Fifth, most of the involved sibling interventions are complex and have multiple components, but none of the study designs allows identification of the most effective components of the intervention. And finally, only two studies (Barrera et al., 2004; Sidhu et al., 2006) conducted follow-up assessment for evidence of the consistency of the intervention effect. However, long term follow-ups would be useful to see whether the benefits remained after termination of the program.
Concerning intervention timing, further issues should be raised. Many of the studies used a broad inclusion criterion for the length of time since onset of cancer or do not take into account time since diagnosis at all. This way interventions can not be tailored to needs and stressors associated with particular stages of treatment (initial time period after diagnosis, end of treatment, relapse, and so on). Regarding intervention documentation, several of the reviewed studies did not describe their interventions sufficiently (for example, detailed program content, theoretical basis, existence of a manual, or lead professional are not specified). This lack of documentation limits the possibility of replication and comparability.

5.3 Directions for future research

Based on the results of this review several recommendations can be made regarding future research in this field. Intervention studies should increase in their methodological quality and be conducted in randomized, controlled designs. To overcome the problem of small sample sizes more multisite collaborative studies should be conducted. Sibling programs should be

manualized and theory based. Relationships among the dependent variables and covariates (age, gender, diagnosis, for example) should be examined further, and there is a need to distinguish content components of the intervention and evaluate them apart. Finally, direct comparison between different settings (individual vs. group), their psychological outcome, and their cost-effectiveness are needed.

In sum this review showed tentative evidence that psychological interventions with siblings of childhood cancer patients can effectively reduce psychological maladjustment and improve medical knowledge about cancer. However, detailed recommendations regarding most effective intervention objectives or settings cannot yet be made.

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III. How siblings of pediatric cancer patients experience the first time after diagnosis: a qualitative study

1. Abstract

Background: Siblings of pediatric cancer patients have a higher risk of developing emotional, behavioral, and social problems. However, little is known about specific experiences of this population in the first time after diagnosis.
Objective: The purpose of this qualitative study was to describe the experiences of siblings of pediatric cancer patients in different areas of life in the first half-year after the cancer diagnosis.
Methods: Semi-structured interviews were conducted with 7 siblings of pediatric cancer patients (ages 11 – 18 years). Siblings were asked about their experiences in the hospital, school, family, and with peers and with the ill child. Content analysis was used to derive important themes from the interviews.
Results: Twenty-two categories of siblings' experiences were identified from the data.
Conclusions: In all areas of life siblings reported difficulties, such as absence of parents, dealing with the ill child's or other patients' suffering and appearance, or impaired school achievement. But the siblings also mentioned important resources such as peer relationship, helpful coping strategies, and increased family cohesion.
Implications: The results of the present study lead to a list of important topics in different areas of life that might be helpful for health care professionals to have in mind when meeting with siblings of cancer patients. Integration of these findings should serve to improve sibling support and develop standardized sibling interventions.

2. Introduction

Whenever a child is diagnosed with cancer, the whole family is affected. With the need to care for the ill child, the entire family is forced to change daily life and routines. Parent focus their attention on the child diagnosed with cancer; healthy siblings are confronted with decreased physical and emotional availability of their parents (Alderfer et al., 2010). Siblings may also worry about the illness, and they have to observe their brother or sister undergo emotional and physical pain. These challenges may be particularly stressful during periods when the ill child is experiencing increased physical symptoms (Woodgate, 2006).
In empirical research on siblings' psychological adjustment, several researchers found that siblings do not experience elevated rates of psychiatric disorders (Alderfer & Hodges, 2010).

However, a significant subset of siblings showed negative emotional reactions like fear, sadness, helplessness, anger, or guilt as well as posttraumatic stress and poor quality of life (Alderfer et al., 2010; Houtzager, Grootenhuis, Caron, & Last, 2004).

To prevent the apparently at-risk population of siblings of cancer patients from developing severe psychosocial problems, early psychological interventions are needed. To design the interventions, it is important to understand the social, emotional, and behavioral consequences of childhood cancer for healthy siblings (Alderfer & Noll, 2006) and to have detailed information about specific experiences in the first months after diagnosis. Several studies examined experiences of siblings of children with cancer, mainly using qualitative methods (Nolbris, Enskar, & Hellstrom, 2007; Sloper, 2000; Woodgate, 2006). The studies highlighted the loss of attention, status, routines, and security; changes in family life; and feelings of concern and anxiety. Positive effects reported were closer family relationships and gains in maturity and compassion. Qualitative research methods seem particularly suitable to identify experiences and needs of siblings, because the design allows siblings to tell their stories in their own words with their own emphases.

Although there is a growing body of knowledge on how siblings experience childhood cancer, most qualitative studies up to now examined siblings' experiences more in general or focused mainly on family and illness issues. Topics that may be important to siblings, such as peers, school, and experiences in the hospital, have not been assessed so far. This study is also the first study to focus on experiences in this very early time after diagnosis, a time period characterized by learning about the diagnosis, the start of medical treatment, and first hospitalizations of the ill child. The aim of this study was to gain an understanding of the everyday life experience of being a sibling when a brother or sister is diagnosed with cancer. To this end we explored qualitatively all the relevant areas of siblings' lives using an in-depth survey of experiences in the domains hospital, school, peers, family, and ill child. Our goal was to obtain an overview of important issues in each area of life and to identify specific difficulties, coping strategies, and possible gains in the first half-year after the cancer diagnosis.

3. Method

3.1 Procedure

Siblings of pediatric cancer patients treated at University Children's Hospital Zurich who met the following eligibility criteria were contacted by mail and invited to participate in the study: (1) brother or sister with cancer diagnosis, (2) age 10–18 years, (3) time since diagnosis between 6 months and 2 years, (4) fluent in German. The lower age limit was set at 10 years to make sure that participants were old enough to reflect upon and express what they experienced. The time lapse of 6 months to 2 years since diagnosis was chosen in order to be sure that the ill child would no longer be in the intensive phase of medical treatment. Children and parents received detailed written information about the study and gave written informed consent. A clinical psychologist conducted the interviews at the participants' homes.

Siblings were interviewed at 8 to 23 months post-diagnosis (mean 16.8 months). Notably, all siblings reacted positively to being interviewed. They appreciated the interviewer's interest and being asked to relate their experiences in detail. All interviews were tape recorded and were from 30 to 80 minutes long. Length of interview was not correlated with age. The local research ethics committee approved the study.

3.2 Participants

Fourteen siblings were contacted; 3 refused participation; 4 did not reply. Seven siblings (all from different families) agreed to participate. All patients were off initial treatment. Table 5 shows demographic variables of the participants.

Table 5. *Demographic variables of the participants*

No.	Sex of sibling	Age of sibling, in years	Age of patient, in years	Rank of the sibling	Months since diagnosis	Sex of patient	Diagnosis
1	F	18	15	1 of 2	21	M	Hodgkin's lymphoma
2	F	18	16	1 of 2	16	M	Acute myeloid leukemia
3	M	13	13	1 of 4	21	M	Acute lymphatic leukemia
4	F	12	12	twin of 2	12	F	Synovial sarcoma
5	M	11	9	1 of 2	8	F	Malignant glioma
6	F	13	6	1 of 3	23	F	Acute lymphatic leukemia
7	F	16	8	1 of 3	17	M	Acute lymphatic leukemia
Mean		14.4	11.3		16.8		

3.3 Interview

We developed an interview guide to assess experiences of siblings after the cancer diagnosis in a qualitative manner. The semi-structured interview focused on five area of siblings' daily life: hospital, school, peers, family life, and ill child. According to clinical experience and previous research, these are relevant areas of siblings' life and are affected by the cancer diagnosis. The following describes the five domains in brief:

1. The *hospital* is most often a whole new environment for siblings. In this part of the interview siblings could express what they experienced while visiting the ill child in the hospital and meeting hospital staff and other patients. 2. The healthy children usually continue to follow daily routines like going to *school*. In this part of the interview siblings reported experiences they had in school after the diagnosis. 3. Another important area of life in school-aged

children is the *peer group*. Peers increasingly influence children's view of themselves and their self-esteem (Murray, 2000). Here siblings were asked about their experiences with peers after the diagnosis. 4. From birth to late adolescence the *family* is primarily responsible for nurturance and protection of a child (Gass, Jenkins, & Dunn, 2007). But childhood cancer impacts the entire family (Kazak et al., 1999). In this part of the interview siblings reported what happened in the family after the diagnosis. 5. Siblings were asked about experiences with the *ill child*. Sibling relationships are usually the longest in people's lives, and siblings share family history and environment (Cicirelli, 1995). With the diagnosis of cancer, both the ill and the healthy child are forced to have new experiences with each other.

Within each of these domains of life, siblings were asked open-ended questions about their experience in general (e.g., "What did you experience in school during this time?") difficulties ("What was difficult?"), and support ("What helped?"). They were encouraged to speak openly. Since our age criterion was quite wide, we took care to use age-appropriate language and communication. The interviewer requested each sibling to recall the first half-year after the onset of cancer and to recall their experiences at that time. A time line was drawn as a visual aid to help the young people stay within the timeframe while answering the questions.

3.4 Analysis

Analyses were carried out using the computer software ATLAS.ti 5.2 (www.atlasti.com). All interviews were first transcribed and then coded by the first author. Content analysis following Mayring (2008) was applied to interpret the results and to identify frequent and notable topics. Whereas the domains of life were formulated a priori based on theoretical aspects, the categories of experience within each area of life were created using inductive category development based closely on the transcripted material. This analysis consisted of the following steps: Units of analysis – that is, the minimal and the maximal part of an interview that may be subsumed under a particular code – were determined. The interviews were read step by step, and the data material was reduced by paraphrasing siblings' statements and assigning codes to units with similar meanings. Statements could be allocated to different codes, as they could refer to several aspects at a time. Further, a provisional category system was constructed by assigning the codes to categories. This category system was refined from interview to interview, until congruence with the complete amount of data was reached. A clinical psychologist reviewed the transcripts to confirm that the categories were comprehensive and reproducible.

A total of 247 units of analysis were extracted from the interview transcripts with a range of 12–51 units per sibling. Twenty-two categories of experiences mentioned by at least 2 siblings were identified from 181 units. The remaining 66 units were statements by one single sibling. Each domain of life covered three to six categories.

Cohen's kappa coefficient (Cohen, 1968) was applied to test the interrater reliability of the categorization. Two clinical psychologists unfamiliar with the categories and the analyses were given minimal instructions on how to assign units to categories. They then categorized

a random sample of 50 units (20 % of all units). Agreement between the original coding and the second coding was almost perfect, with a kappa of 0.82 in both cases (Landis & Koch, 1977).

4. Results

Siblings' responses could be differentiated into 22 categories, to which in each case at least 2 siblings had contributed. Table 6 shows the categories in all five domains of life.

4.1 Hospital

Taken together, healthy siblings participate in what is going on in the hospital. Siblings mostly want to be on site, they get in touch with other patients and hospital staff and they appreciate being involved in the care of their ill brother or sister.

Other patients: Regarding hospital experiences, being aware of or in contact with other patients besides the ill family member seemed to be important for a majority of siblings. They saw other patients with a changed appearance and witnessed their suffering. They also realized that other patients died of cancer. Besides these distressing experiences, siblings also became acquainted with other children in the hospital, and they even kept up contact when their ill brother or sister's therapy ended.

> *"They talked again and again about children who died. That was sad, even though I did not know the ones who died." (No. 7)*
>
> *"I was always a bit sad when I went to the hospital, because I saw other children there in very bad condition." (No. 6)*

Assistance: Four siblings reported being actively involved in the care of the hospitalized child. They brought food to the hospital, read aloud to the ill child, and assisted with daily routines. Being present, helpful, and part of the process was important for most of the siblings. But they also mentioned that it was sometimes difficult for them to find out when and how to help.

> *"Well, I always asked if I should help. I never quite understood when to help and when I'd better not." (No. 5)*

Reality check: Most of the siblings described the hospital visits as a good opportunity to become familiar with the hospital environment and to form a realistic picture of the medical condition of the ill child. Siblings' preexisting and often negative picture of hospital could be revised, and being on-site helped them to cope better with the situation.

> *"I thought it was not so nice there in the hospital, since my brother always said he couldn't stand going there. But actually it's not so bad once you're there." (No. 1)*

Table 6. *Domains of life and categories of experiences (n = 7)*

Domains of life	Categories	n
1. Hospital	Other patients	5
	Assistance	4
	Reality check	4
	Contact with hospital staff	4
2. School	Academic achievement	6
	School aversion	2
	Questions at school	2
3. Peers	Distraction	4
	Talk about the illness with peers	4
	Questions from peers	3
	Reduced social activities	2
	Normality	2
4. Family life	Absence of parents	7
	Family cohesion	6
	Talk about the illness in the family	5
	Household duties	5
	Restrictions on holidays	3
	Impairment of parents	3
5. Ill child	Ill child's appearance and suffering	7
	Behavior changes	5
	Fear and worries	3
	Jealousy	3

Note: The numbers represent the number of participants who gave responses in a category.

Contact with hospital staff: Many siblings appreciated personal contact with physicians and nurses at the hospital. They described hospital staff as friendly, helpful, and willing to inform them about the illness. One sibling even got help on homework from a ward nurse.

> *"The nurses were nice and all. They talked to me a lot, about how my brother was doing, and what he had to do, and stuff like that." (No. 2)*

4.2 School

At first glance, school may be seen as a part of siblings' life that is not immediately affected by the cancer. However, as the data show, siblings are confronted with several difficulties in

school due to the illness, especially in the first time after diagnosis.

Academic achievement: Almost all siblings reported problems in academic achievement in the first time period after cancer diagnosis. The main problem was being distracted by thoughts and worries about the ill child's condition and life. As a result, the healthy child had trouble with concentration during class and homework, and 3 of 6 siblings even had declines in their grades. Yet, the problems seemed to be temporary. In 5 of 6 siblings, academic achievement returned to normal after a couple of months.

> *"I couldn't follow my classes, and I was distracted a lot. I was always thinking about something else; I worried about my brother." (No. 2)*
>
> *"Well, it was really bad at the beginning. I couldn't concentrate at school, I had headaches in class, or I felt queasy. But after two, three months things started to get better. Especially after the first report from the hospital that the tests now showed nothing." (No. 4)*

School aversion: Due to the changed situation at home, some siblings had a strong wish to be absent from school and to stay with and support the ill child.

> *"When my sister had to do something, I could never be there. I would rather have gone with her to the hospital instead of to school. But when she wasn't well, I always had to go to school after all." (No. 6)*

Questions at school: At school siblings were frequently asked about the ill child's condition and diagnosis. Two siblings stated that questions from teachers and classmates made them feel uncomfortable. To be the informant was fine when they wanted to volunteer the information but not when they were forced to report. The two siblings each found a helpful strategy to avoid this conflict: One referred inquiries to a friend at school who was informed about the ill child's condition; the other set up a website that gave everyone access to the latest news on his sister's condition.

> *"Well, everyone at school came to me and asked how my brother was doing. Even the teachers kept asking. And after a while I thought, why do they always have to come and ask me?" (No. 7)*

4.3 Peers

Peers are an important source of support for siblings, distracting them from the difficult situation at home. However, the cancer of the ill child can also decrease opportunities to maintain these significant relationships.

Distraction: Interaction with peers helped provide a majority of siblings with some distraction from the illness issues. Spending time with peers and discussing peer group topics made them forget their worries about the ill child.

> *"My friends helped me; I could get away from it all when I spent time with them, and I could forget about the illness for a moment. I could be a little happy with them." (No. 6)*

Talk about the illness with peers: Four siblings described talking to their friends about cancer and the ill child as a positive experience. Conversation provided feelings of relief, comfort, and hope.

> *"Actually it was good to talk about it with them. You get rid of a burden somehow." (No. 1)*

Questions from peers: As at school, siblings had to answer many questions from peers about the illness and the ill child's condition. The 3 siblings reporting this issue felt bothered by questions from peers, especially when they were asked the same things over and over and when they were asked how the ill child was and never how they themselves were.

> *"They kept asking me if he will survive and things like that. They always asked me the same things" (No. 2).*

Reduced social activities: Some siblings reported decreased social activities in the first months after the diagnosis. Spending a lot of time at the hospital with the ill child was one reason why they could not keep in touch with friends. Another sibling did not feel like going out with friends while their brother or sister was in a bad condition. And after having been away for a while, it was difficult to reintegrate into the group.

> *"You don't know much about what's going on because you can't get together with your friends as often. And when you meet again, at first you don't know what to talk about. And they don't know what to talk about with you, either." (No. 2)*

Normality: Some siblings particularly appreciated the fact that their friends did not act differently after the cancer diagnosis and maintained a sense of normality.

> *"Actually my friends stayed the way they were before. I was hoping it would be that way." (No. 5)*

4.4 Family life

Changes in family life are a crucial experience for siblings of children with cancer. Family life is characterized by physical separation of family members and by heavily burdened parents, changed daily routines, as well as increased closeness between family members.

Absence of parents: An important topic mentioned by all siblings interviewed was parents' absence in the first months after the diagnosis. Siblings experienced physical separation from parents caused by hospitalization of the ill child. Typically, one parent stayed at the hospital permanently, and the other parent spent considerable time there on visits. Most siblings felt lonely and disliked their empty home. Parents were not available for important things like helping with homework, listening to what was going on in the sibling's life, and having meals together. In the majority of cases separation from parents was a burden. Two siblings merely noticed their parents' absence without having negative feelings, and 1 even liked being alone at home from time to time.

> *"Before they came home for the first time, something was missing. It was empty. It was quiet, even when the radio was on, for me it was quiet. […] My mom used to work in the morning and at lunchtime she went to the hospital. And my father went to the hospital right after work because he works in town. Then, of course, they stayed there for quite a while, and they usually didn't get home until ten o'clock in the evening." (No. 1)*
>
> *"The most difficult thing was that my parents were away so often. I was alone at home. […] Sometimes I didn't see my mom for a whole week. I only saw her when I went to the hospital." (No. 6)*

Family cohesion: Alongside all the difficulties, 6 of 7 siblings reported increased cohesion and closeness between family members as a result of the cancer diagnosis. They described how the family held together, how they got along better with the ill child and other siblings, and how arguments in the family were rare during this time.

> *"My brother and I got much closer in this time, and we spent more time together than before." (No. 7)*
>
> *"I spent much more time with my father in those days than with my mother. I realized he was there for me, too, when I wasn't doing well. He kept asking me how I was doing, and I liked that." (No. 2)*

Talk about the illness in the family: Five siblings reported that illness and the patient's health condition were constantly discussed in the family as a dominating topic of conversation. Siblings generally appreciated being informed about the ill child's state of health, but they were also tired of talking about nothing but the illness with other family members.

> *"We didn't talk about anything but the illness. There was nothing else we talked about." (No. 1)*
>
> *"Well, at dinner we talked a lot about her illness and that was… Actually, I didn't like that at all." (No. 4)*

Household duties: Five siblings reported that they took over more household duties, such as cooking, cleaning, or doing the laundry. Although they did not do this voluntarily, none of them complained about having additional chores. For some siblings, taking over these chores was even a welcome opportunity to help out their parents and show cooperation.

> *"I had to do the laundry quite often. I also cooked for my brother or my father and cleaned the kitchen afterwards. I had to keep house a bit. Actually, I liked doing it." (No. 7)*
>
> *"I cleaned up a lot, in all the rooms, not only in mine. And I had to look after my youngest brother quite often." (No. 6)*

Restrictions on holidays: Another loss of family routines experienced by 3 siblings concerned restrictions on holidays. Vacations planned before the cancer diagnosis had to be canceled, or siblings had to spend holidays with relatives instead of parents. Siblings had to stay at home more, even though a change of scene would have been a welcome distraction.

> *"We had lots of plans for traveling that year. We couldn't do it. I had really been looking forward to it, and so I was very disappointed when we couldn't go abroad." (No. 6)*

Impairment of parents: Several siblings reported that their parents were mentally distressed and psychologically impaired. To witness their parents being burdened, weak, sensitive, or irritable was difficult for siblings and resulted in some cases in role changes (they wished to protect and comfort parents).

> *"I noticed she wasn't well. You could tell, because she talked about the illness all the time. And she was physically fragile. And she was always under stress … Well, it bothers you, because you don't know your parents like that, that they can be so weak." (No. 1)*

4.5 Ill child

Siblings are very much aware of the consequences of the illness and the treatment for the ill child. They observe their brothers' or sisters' appearance, suffering, behavior change as well as their being the center of attention. Siblings react with a wide spectrum of emotions, including fear and jealousy.

Ill child's appearance and suffering: When asked about experiences with their ill brother or sister, all siblings reported witnessing suffering and changes in appearance in the ill child. They had to watch their brothers or sisters losing their hair, gaining or losing weight; they saw them connected to medical equipment and experiencing pain. All these observations led to negative emotions. Siblings felt helpless and sorry for the ill child; they were shocked, had feelings of disgust, and felt unsure of how to react.

> *"Overnight he suddenly had no hair. That was terrifying for me. And then I also didn't know what to say not to hurt him. Finally, I just tried to comfort him." (No. 2)*
>
> *"He looked strange. When he was given Cortisone he ate a lot. Then he didn't eat for a while. Then he had no hair; now he has lots again. In pictures from those days you wouldn't recognize him." (No. 3)*

Behavior change: Behavior changes in the ill child were also a relevant experience for a majority of siblings. They noticed their sister or brother being more aggressive, moody, or passive, and witnessed them having all kinds of special requests. Two siblings were able to connect this behavioral change with side effects of medications. For all of them, dealing with these new behaviors was difficult.

> *"He was demanding. Sometimes he wanted sausages with ketchup and all sorts of things at one o'clock in the morning, for breakfast spaghetti – all confused. And during a certain phase he got aggressive very fast." (No. 3)*
>
> *"It was because of the meds. When we went on vacation she was all angry, and the vacations weren't exactly nice for me. She always wanted some particular thing, and she was furious all the time." (No. 6)*

Fear and worries: Regarding the severity of the diagnosis, 3 siblings reported having deep sorrow and fear about the prognosis of their sister or brother's disease. They worried about efficacy of treatment, feared an additional diagnosis, and feared the sick child's death.

> *"The moment it came out [diagnosis], I could only think of the fact that my brother could die from the illness." (No. 2)*

Jealousy: Three siblings talked about jealousy during this time period, reporting being jealous of the attention and gifts that the ill sibling received. And even though they understood why attention was unequally distributed, their jealousy was difficult to suppress. One sibling described the reverse situation: His sick brother was jealous of him, because he was healthy and not as restricted in his daily living.

> *"My sister was, and I actually do understand this, showered with gifts, really showered with them. And I thought I might also be glad of a little cuddly toy or whatever. I understood all this, of course – but still, they treated me as if I wasn't there sometimes." (No. 4)*

5. Discussion

In this study we used a qualitative research method to analyze interviews with siblings with the aim of collecting more information about their experiences in the first six months after their sister or brother was diagnosed with cancer. This study was the first to reveal issues in all relevant areas of siblings' lives, namely, hospital, school, peers, family life, and ill child, in this early time after diagnosis. The interviews conducted revealed 22 categories of siblings' experiences across all five domains of life. These experiences included several difficulties that siblings had to manage, but the siblings also developed resources and coping strategies and obtained gains in this time period.

Difficulties occurred in all areas of life. Siblings in our study described how family life after the cancer diagnosis was determined by the ill child's treatment protocol, resulting in their parents' regular absence and reduced family activities, a fact that also other qualitative studies in this research area reported (Freeman, O'Dell, & Meola, 2000; Sloper, 2000; Woodgate, 2006). Illness and treatment also dominated conversation in the family, all in all leaving the healthy siblings feeling that they were not getting any attention and feeling lonely. Previous reviews of siblings' adjustment also found these changes in family functioning and consequences for siblings (Alderfer et al., 2010; Wilkins & Woodgate, 2005). But besides the reduced availability of their parents, healthy siblings also found it difficult to handle the psychological impairment of mothers and fathers, a topic hardly touched upon in previous reports.

Living together with a sick brother or sister also resulted in sibling jealousy and evoked intrusive worries about the ill child's health and prognosis, a finding that was previously reported (Nolbris et al., 2007; Patterson, Holm, & Gurney, 2004; Sidhu, Passmore, & Baker, 2005; Sloper, 2000; Woodgate, 2006). Interviewed siblings also expressed worries about their brother or sister's altered behavior, and it was difficult for all siblings to observe the ill

child's changed appearance. Nolbris and Hellstrom (2005) and Scott-Findlay and Chalmers (2001) previously reported similar findings. Besides the family member hospitalized, siblings reported being affected also by other patients in the hospital, a finding that was not previously described in the literature. Witnessing other ill children suffering, observing their altered appearance, or even hearing about the death of a patient was difficult for siblings. Further difficulties emerged at school, with siblings reporting a decline in academic performance, a problem also reported by Fife, Norton and Groom (1987) and Heiney et al. (1990), and an aversion to take part in everyday school life during the time they felt the need to support the ill child. Fielding questions was seen as a difficult task by siblings at school and in the peer environment. As the ones in the family going to school regularly and being out and about in the neighborhood, siblings are probably the family members who are confronted the most often with others' curiosity and questions. And they are not always in the mood to answer, because the subject is burdensome, they are tired of repeating the same answers, or they do not know the answers themselves. We did not find any other studies referring to this issue.

The interviews further revealed that siblings' peer activities were reduced and difficult to maintain due to the changed situation in the family. This seems particularly unfavorable, since peer relations contributed largely to improvements in siblings' psychological state (Alderfer & Hodges, 2010). This finding of reduced social activities is consistent with results of earlier studies (Sidhu et al., 2005; Sloper, 2000).

However, not all experiences of siblings were negative. Siblings' accounts indicated that supportive relationships with peers were *resources* that helped them cope with the situation. Peers allowed siblings to talk about their situation, to enjoy some welcome distraction, and to experience normal life away from the situation of serious illness in the family. Other partners, including hospital staff or other inpatients, also supported siblings in a positive way. The importance of supportive relationships was documented in earlier studies (Alderfer & Hodges, 2010; Nolbris & Hellstrom, 2005; Sloper, 2000). Further, siblings identified a number of *coping strategies* that they felt helped them deal with their situation. Being actively involved and helping in some way, by taking part in the ill child's care or taking over household chores, made siblings feel valuable and decreased their helplessness. Siblings also realized that spending time with the ill child and becoming familiar with the hospital environment helped them to form a real picture of the situation. Active involvement in the treatment process was found to be important for siblings in earlier studies (Barrera, 2000; Kramer, 1984; Sloper, 2000). Finally, under the stress of all the questions about diagnosis and treatment from outsiders like teachers, classmates, or even friends, siblings developed unique coping strategies to relieve the burden by referring people to a special website or to other informants.

Beyond the difficulties that siblings are confronted with, our study revealed *gains* in terms of improved family relationships and cohesion. Siblings reported increased closeness and a strengthening of bonds between family members after the experience of acute illness in the family. This positive response is consistent with previous research on siblings', patients', and parents' adjustment following childhood cancer (Barakat, Alderfer, & Kazak, 2006; Chesler, 1991; Sargent et al., 1995).

5.1 Strengths and limitations of the study

The strength of this study is the inclusion of siblings themselves rather than reports from parents or health care professionals. The qualitative nature of the design allowed siblings to report thoughts and concerns without being restricted by others' beliefs and yielded rich detail on a range of experiences of siblings of childhood cancer patients. But this study has several limitations. First, the number of siblings interviewed was small. Some issues, only reported by one sibling, were not mentioned in the report but might be important considering a larger group. Second, unintentionally all siblings in this sample were older than the ill child. We cannot exclude that siblings younger in birth rank would have reported different experiences. And third, the study was retrospective and cross-sectional in design, so reports were limited to what siblings remembered and wished to share about their experiences. Time lapse since diagnosis was quite large in some cases and made thinking back quite challenging. In future, research with larger and more representative samples is desirable. In addition, a more complete picture of siblings' experiences might be obtained through serial interviews conducted at specific time points after the diagnosis.

5.2 Implications for Practice

These findings provide health care professionals with some information that may be of help in understanding the situation of siblings in the first months after diagnosis and in providing appropriate care. By assessing experiences in all relevant domains of their lives we created a list of topics that might be helpful to have in mind when meeting with siblings of cancer patients. Considering that healthy siblings benefit from being involved in the cancer treatment regime, it seems important to facilitate the involvement of siblings in the care of their ill brother or sister, as long as it is appropriate and not overly demanding (Alderfer et al., 2010). Parents should be encouraged to bring siblings to the hospital and to have them meet with the hospital staff (Spinetta, 1999). Further, health care professionals should ensure that information and support is available for siblings and should call parents' attention to siblings' needs and possible reactions in several domains of life, including difficulties at school, in the peer group, and in the family. Possibly, siblings should be encouraged to not feel obliged to answer all questions about illness and treatment.

This study adds to the evidence in the literature of the challenges siblings face in pediatric oncology and should aid development of psychological intervention protocols for siblings. Our findings and results from previous studies show clearly that siblings' distress is substantial in the first months after diagnosis and that interventions should be provided early (Ballard, 2004; Lahteenmaki, Sjoblom, Korhonen, & Salmi, 2004). The cancer diagnosis affects most domains of siblings' daily life; therefore, psychological interventions should take into account several areas. Besides providing siblings with information about the hospital and illness-related issues, siblings should be supported in family-related problems and difficulties at school and in the peer environment. To give a sibling a more global picture of their situation and to normalize their feelings, professionals can speak to them of the difficulties expe-

rienced by other siblings and about the coping strategies they use. At all times, it is important that siblings be shown appreciation and given praise for what they contribute in their family.

5.3 Conclusions

This qualitative study reported siblings' own experiences in their daily lives during the first time period after diagnosis, thus contributing to new knowledge in this field. The findings revealed siblings' differentiated experiences, for the young people reported both difficulties and perceived positive changes and resources. With a better understanding of the situation of siblings, health care professionals and parents can provide useful information and effective support, helping healthy siblings to cope successfully with the cancer of their brother and sister.

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IV. A Two-session psychological intervention for siblings of pediatric cancer patients: a randomized controlled trial

1. Abstract

Background: Since siblings of pediatric cancer patients are at risk for emotional, behavioral, and social problems, there is considerable interest in development of early psychological interventions. This paper aimed at evaluating the effectiveness of a two-session psychological intervention for siblings of newly diagnosed pediatric cancer patients.
Procedure: Thirty children age 6–17 years were randomly assigned to an intervention or control group. The manualized intervention provided to siblings in the first 2 months after the cancer diagnosis of the ill child included medical information, promotion of coping skills, and a psycho-educational booklet for parents. At 4 to 6 weeks, 4 months, and 7 months after the diagnosis, siblings and their parents completed measures (from standardized instruments) of social support, quality of life, medical knowledge, posttraumatic stress symptoms, and anxiety.
Results: At follow-up siblings in the intervention group showed better psychological well-being, had better medical knowledge, and reported receiving social support from more people. However, the intervention had no effects on posttraumatic stress symptoms and anxiety.
Conclusions: These initial findings suggest that a two-session sibling intervention can improve siblings' adjustment, particularly psychological well-being, in the early stage after a cancer diagnosis.

2. Introduction

Having a brother or sister newly diagnosed with cancer is a distressing and challenging situation for the healthy child. The cancer diagnosis in the family has emotional, behavioral and social consequences for healthy siblings (Alderfer et al., 2010). Siblings are confronted with changed daily routines in the family and decreased physical and emotional availability of their parents (Alderfer et al., 2010; Sloper, 2000). They are worried about the illness and have to observe their brother or sister have emotional and physical pain. These experiences may lead to intrusive and conflicting emotions such as fear, loneliness, sadness, anger, jealousy, or guilt (Bendor, 1990; Nolbris, Enskar, & Hellstrom, 2007; Sidhu, Passmore, & Baker, 2005; Sloper, 2000; Woodgate, 2006).
Previous research on adjustment of siblings of children with cancer found most siblings' general adjustment to be within normal limits (Alderfer et al., 2010). However, a significant

subset of siblings suffers from cancer-related posttraumatic stress symptoms (PTSS) (Alderfer, Labay, & Kazak, 2003; Alderfer et al., 2010; Packman, 2004), and there is evidence of poorer health-related quality of life (HRQoL) in this population (Houtzager, Grootenhuis, Caron, & Last, 2004; Houtzager, Grootenhuis, Hoekstra-Weebers, & Last, 2005; Packman et al., 2005). School and social functioning may be impaired in the first time period after diagnosis (Fife, Norton, & Groom, 1987; Labay & Walco, 2004; Lahteenmaki, Sjoblom, Korhonen, & Salmi, 2004). In sum, the findings on psychosocial adjustment of siblings of pediatric cancer patients indicate that siblings do not suffer from severe psychopathology but are at risk for emotional, behavioral, and social problems, typically soon after the diagnosis (Alderfer et al., 2010; Houtzager, Grootenhuis, & Last, 1999).

Given the difficult circumstances that childhood cancer causes for all family members, it is important to understand the consequences of the diagnosis for siblings and to develop appropriate interventions to reduce distress and promote psychological adjustment. It is important to identify the needs of siblings and to recommend and encourage appropriate treatments when problems are detected (Alderfer & Noll, 2006). However, intervention research with siblings is still rare (Kazak, 2002). The majority of published papers reported results of descriptive and correlational research, and there is a lot of non-empirical, anecdotal data. A previous review of empirically evaluated intervention programs showed improvements in siblings' depression symptomatology, medical knowledge about cancer, HRQoL, and high satisfaction ratings in siblings and parents (Prchal & Landolt, 2009). But many existing studies are methodologically weak; most intervention studies relied on simple pre/post evaluations and only a minority used randomized controlled designs (RCTs) (Prchal & Landolt, 2009). Concerning intervention timing, most studies used a broad inclusion criterion for the length of time since onset of cancer. Only one intervention so far reported targeting siblings of patients in an early stage of treatment and therefore aimed at prevention (Sidhu, Passmore, & Baker, 2006). To our knowledge all published papers on empirically evaluated interventions with siblings used interventions in a group or camp format.

The present study aimed at assessing the effect of a two-session early psychological intervention for siblings of pediatric cancer patients using a randomized controlled trial. This intervention was provided early, i.e., within the first two months of diagnosis, and was conducted in an individual format. We expected this intervention to effectively reduce anxiety and PTSS and to improve medical knowledge, social support, and HRQoL of siblings.

3. Method

3.1 Participants

Participants were recruited at two hospitals in Switzerland: University Children's Hospital Zurich and Children's Hospital Lucerne. Siblings had to meet all of the following criteria: (1) brother or sister with newly diagnosed childhood cancer, (2) medical treatment (inpatient or outpatient) necessary, (3) age of healthy sibling from 6 to 17 years, (4) fluency in Ger-

man. Families with a child who met criteria for inclusion were contacted within 1 month of diagnosis. If the family had several healthy siblings who met the inclusion criteria, all willing siblings were included.

Forty-five siblings met the inclusion criteria and were asked to participate. Fifteen siblings and their families declined participation (33.3%); reasons were because the child refused to participate (46.7%), because the study seemed too time-consuming (26.7%), or because parents thought the study would be an additional strain on the family (26.7%). Due to incomplete data at follow-up assessments (one drop-out), the final study sample comprised 29 children (response rate 64.4%) from 21 families (see Figure 4). Comparison of participants and non-participants revealed no significant differences in mean age of healthy siblings ($t = 0.47$, $p = .64$), sex ($\chi^2 = 0.72$, $p = .40$), type of diagnosis ($\chi^2 = 0.88$, $p = .64$), intensity of treatment ($Z = -0.31$, $p = .76$), medical complications ($Z = -0.22$, $p = .83$), and health-related restrictions ($Z = -0.50$, $p = .62$).

A total sample size of 32 cases (16 for each group) would be required to detect a statistically significant effect size of 0.9 in a single tail t-test with a power of 0.80 and Type I error rate set to .05. Expected effect size was based on previous studies on sibling interventions (Prchal & Landolt, 2009) and on the notion that an individual intervention would achieve slightly higher effects.

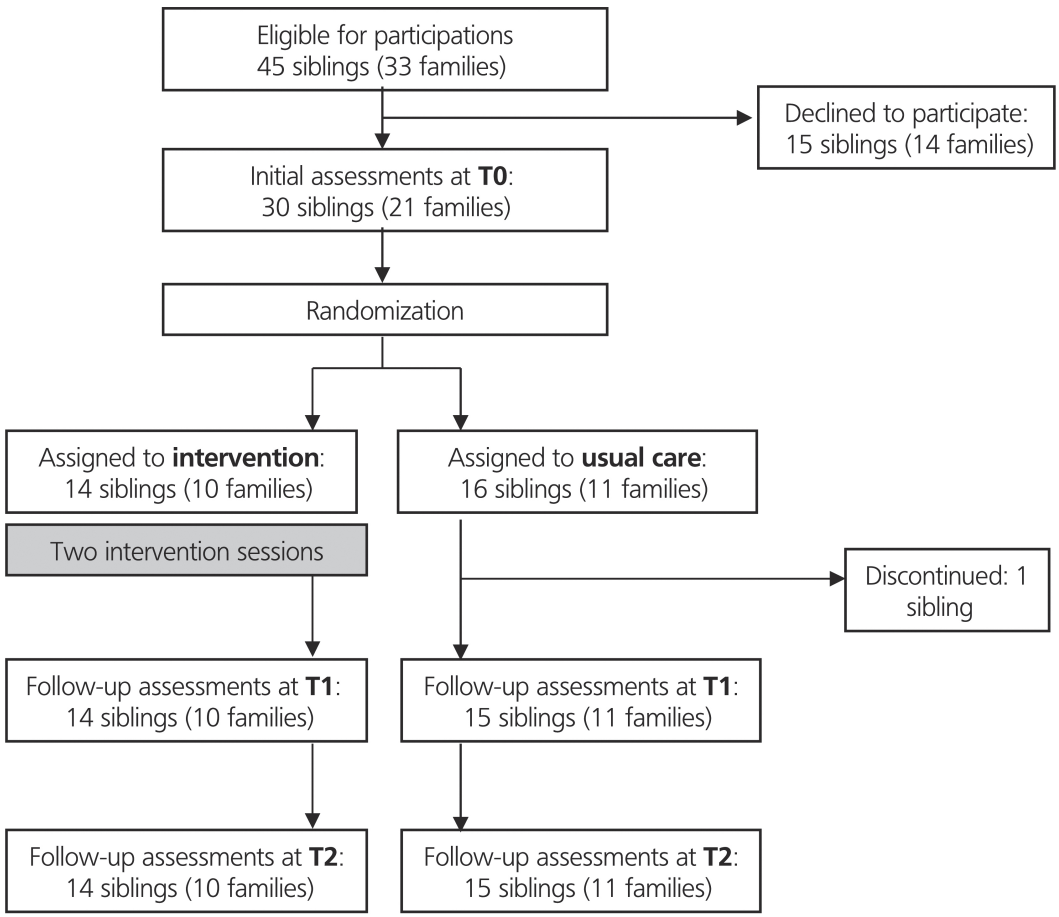

Figure 4. *Diagram of the study cohort.*

3.2 Procedure

The institutional review boards of both study sites approved the study. Written informed consent was obtained from parents and siblings older than 12 years. Assessments were carried out at 4 to 6 weeks (T0; M = 36.0 days, SD = 10.2 days), 4 months (T1; M = 132.9 days, SD = 16.9 days), and 7 months (T2; M = 220.9 days, SD = 16.6 days) after the cancer diagnosis. The siblings were assessed by means of standardized interviews conducted by trained psychologists. The interviews lasted approximately 45 minutes, and most of them were conducted in the siblings' home; some were conducted at the hospital. Mothers and fathers were independently assessed at the same time using questionnaires. Medical variables were retrieved from the responsible physicians. In return for participation, families received 50 Swiss francs after completing all three assessments.

The randomization list was generated by the program Rancode 3.6 (IDV, Gauting, Germany) at the beginning of the project and stratified for sex. Participating siblings were randomly assigned to the intervention or the control group. If several siblings in one family agreed to participate, the sibling closest in age to the ill child was randomly assigned according to sex and then all siblings of the family attended the same study group.

If the child was assigned to the intervention group, the first session of the intervention was conducted immediately after the T0 baseline assessment. The second intervention session was held 2 weeks later. A different interviewer, who was blind to the child's status in the project, conducted follow-up assessments at T1 and T2 with both control and intervention group.

3.3 Measures

Medical knowledge: Medical knowledge about cancer was assessed using a scale that we developed. Siblings answered four questions: on name of the illness, medical understanding of the illness process, treatment options, and length of treatment. Using a comprehensive coding sheet, two clinical psychologists coded the answers together on a scale ranging from 0 to 2, with higher scores indicating better medical knowledge. Cronbach's alphas for the medical knowledge scores were satisfactory to poor, with $\alpha = 0.63$ at T0, $\alpha = 0.50$ at T1 and $\alpha = 0.34$ at T2.

Social support: The number of individuals providing social support was assessed using a scale that we developed. Siblings received a comprehensive list of individuals that were available to them and might be sources of social support, including mother, father, grandparents, siblings, relatives, godmother and godfather, neighbors, close friends, peers, and teachers. They then indicated whether or not these individuals provided social support. The score was the total number of people providing support.

Health-related quality of life: HRQoL was assessed using KIDSCREEN-27, a standardized questionnaire for children from 8 to 18 years of age (Ravens-Sieberer et al., 2005). The KIDSCREEN instruments were developed and validated in several European countries simultaneously. KIDSCREEN-27 contains 27 items building five subscales: physical well-being, psychological well-being, autonomy and parents, social support and peers, and school environ-

ment. In this study, we used both the child version and the parent version, filled out by mothers. All scores are reported as T-values, based on Swiss community norms, with higher scores indicating higher HRQoL. KIDSCREEN-27 was found to be a reliable and valid measure of HRQoL in children and adolescents (Ravens-Sieberer et al., 2007). In this sample, KIDSCREEN-27 showed very good internal consistency (self-report: $\alpha = 0.85$ at T0, $\alpha = 0.88$ at T1 and $\alpha = 0.89$ at T2; proxy report: $\alpha = 0.87$ at T0, $\alpha = 0.89$ at T1 and $\alpha = 0.90$ at T2).

Posttraumatic stress symptoms: Siblings were interviewed about their illness-related posttraumatic stress symptoms using the UCLA PTSD Reaction Index (UCLA RI) (Steinberg, Brymer, Decker, & Pynoos, 2004). We altered the wording slightly to ensure that siblings reported on their experience of their brother or sister's cancer. The items of the UCLA RI closely follow the DSM-IV symptoms of posttraumatic stress disorder (PTSD) and can provide diagnostic information. Siblings were asked about their reactions during the past month and ranked their responses on a 5-point Likert scale from 0 (none of the time) to 4 (most of the time). An overall score was calculated (range 0-68), with higher scores indicating greater symptom severity. As to internal consistency of the UCLA RI, several reports found Cronbach's alpha to fall in the range of 0.90 (Steinberg et al., 2004). In this sample, Cronbach's alpha was 0.90 at T0, 0.88 at T1, and 0.91 at T2 for the overall score.

Anxiety: The Spence Children's Anxiety Scale (SCAS) (Spence, 1998) is a 44-item self-report questionnaire to assess the severity of anxiety symptoms broadly in line with the DSM-IV dimensions of anxiety. It assesses six domains of anxiety: generalized anxiety, panic and agoraphobia, social phobia, separation anxiety, obsessive-compulsive disorder, and physical injury fears. The addition of all scores results in a total score. In this study we used the German version and norms of the SCAS (Essau, Muris, & Ederer, 2002). Children rated the frequency with which they currently experienced each symptom on 4-point scale, with never (0), sometimes (1), often (2), and always (3). Internal consistency of the SCAS total score was excellent, with a Cronbach's alpha of 0.96 at T0, 0.93 at T1, and 0.94 at T2.

Socio-economic status: Socio-economic status (SES) as assessed by mothers was calculated by means of a 6-point score of both paternal occupation and maternal education. Three social classes were defined as follows: scores 2−5, lower class; scores 6−8, middle class; and scores 9−12, upper class. This measure was used in previous studies and was shown to be a reliable and valid indicator of SES in Switzerland (Landolt, Vollrath, & Ribi, 2002).

Medical variables: The pediatric oncologist in charge was asked to rate the following three medical variables concerning the ill child on a 3-point scale: *Intensity of treatment* (1 = low: surgery only or 6 months' chemotherapy only or both, with favorable prognosis; 2 = medium: treatment longer than 6 months according to the treatment protocol, with intermediate prognosis; 3 = high: treatment according to high risk protocols, bone marrow transplantation, with unfavorable prognosis), *Medical complications* (0 = no complications and good response to therapy; 1 = moderate complications, e.g., hospitalization due to infection; 2 = severe complications, e.g., multiple hospitalizations due to infections, no response to treatment) and *Health-related restrictions* (0 = no restriction; 1 = moderate restrictions, e.g., distinct fatigue, pain; 2 = severe, e.g., intense pain, considerable restrictions in physical and cognitive

performance). The intensity of treatment and the medical complications items were used successfully in previous studies on children with cancer (Landolt, Vollrath, Niggli, Gnehm, & Sennhauser, 2006).

3.4 Intervention

Our intervention was provided within the first two months after the cancer diagnosis and therefore during a stage which has been shown to be the most vulnerable time for siblings' adjustment (Alderfer et al., 2010; Lahteenmaki et al., 2004). Also, by targeting a specific time frame, the intervention can be tailored to needs and stressors associated with this particular initial time period after diagnosis. By choosing an individual as opposed to a group format, we aimed for developmentally appropriate intervention and wanted to allow for flexibility to address personal concerns and provide individualized cancer-related information.
We developed the standardized psychological intervention based on clinical experience, theoretical considerations, the relevant literature, and a qualitative pilot study that had gathered more information about siblings' experiences in this time period (Prchal & Landolt, 2011). In two sessions, each approximately 50 minutes long, a clinical psychologist (first author of this paper) guided siblings and their parents through a three-step program: (1) medical information, (2) coping with stressful situations, and (3) information for parents.
The medical information part focused on the siblings' understanding of body functioning, the illness mechanism and location, and the cancer treatment, in particular chemotherapy. To accomplish this goal, pictures and storybooks were presented as aids. It was particularly emphasized that nobody was to blame for the development of cancer, nobody had done anything wrong, and cancer is not contagious. By learning more about the disease, treatment schedule, and side effects, siblings should gain a feeling of control over the situation which might reduce feelings of anxiety (Last & Grootenhuis, 1998; Sahler & Carpenter, 1989) and enhance social competence (Evans, Stevens, Cushway, & Houghton, 1992).
The part on coping encouraged siblings to think of changes and particular stressful situations in their life since the cancer diagnosis. Three significant stressors were looked at in detail, and helpful coping strategies were discussed. Results of the coping session were written down on a special list and handed out to siblings. At least one parent joined after this part of the intervention. With the sibling's agreement, parents were informed about the relevant topics of the intervention in order to get as much support from parents as possible. Cognitive behavior therapy was the therapeutic approach used during problem identification and discussion of coping strategies. Following the coping and stress model proposed by Lazarus and Folkman (Lazarus & Folkman, 1984) siblings were encouraged to appraise stressors in their daily life and develop coping strategies in response to their specific situation.
In the last part of the intervention, parents received a psycho-educational booklet developed by the authors containing information on the psychosocial situation of siblings of cancer patients in general and providing recommendations to parents on how to support their healthy children. At the end of each session there was time for questions from siblings and parents.

3.5 Control condition

Families in the control group received standard psychosocial care, which consisted of meetings with the psycho-oncologist on the ward, who was primarily responsible for the ill child and the parents but also met with siblings if necessary. After follow-up assessments were completed, the control group was offered individual sessions for siblings.

3.6 Statistical analyses

For data analysis we used statistical package SPSS for Windows, release 16 (SPSS Inc., Chicago, IL). All analyses were performed with two-sided tests. A p value ≦ .05 was considered significant. Kolmogorov-Smirnov Goodness of Fit Tests showed normality for all outcome measures. To compare nominal and ordinal scales, χ^2 analyses and, when cells were too small, Mann-Whitney U tests were used. Normally distributed continuous data were analyzed using independent t-tests. To determine the effectiveness of the intervention, two-factor repeated measures analysis of variance (ANOVA) were performed. In the statistical analysis, siblings' adjustment variables were compared with respect to group, time, and group × time interaction. Post-hoc analysis for significant time effects was corrected for multiple comparisons using Bonferroni adjustment. If significant mean differences were detected, effect sizes (d) were calculated following Cohen (Cohen, 1988).

4. Results

4.1 Sample characteristics and baseline assessment

Table 7 shows sample characteristics. The intervention and the control groups did not differ significantly on any demographic or illness characteristic. Likewise, none of the medical variables showed group differences at any assessment time point: Intensity of treatment (T0: Z = -0.61, p = .54; T1: Z = -0.25, p = .80; T2: Z = -1.08, p = .28), medical complications (T0: Z = 0.00, p = 1.00; T1: Z = -1.29, p = .20; T2: Z = -0.75, p = .45), health-related restrictions (T0: Z = -0.94, p = .35; T1: Z = -0.48, p = .63; T2: Z = -0.72, p = .47). Similarly, there were no significant between-group differences on any baseline outcome measure at T0: medical knowledge: t = 1.32, p = .20; social support: t = 1.17, p = .25; KIDSCREEN self-report: t = 1.26, p = .22; KIDSCREEN mother-report: t = 0.05, p = .96; UCLA PTSD: t = 1.39, p = .18; SCAS: t = 1.12, p = .27.

At baseline the total sample did not differ from community norms on mother-reported HRQoL (t = -1.31, p = .20) and anxiety (t = 0.58, p = .57). But self-reported HRQoL at baseline was significantly lower than in the Swiss norm population (KIDSCREEN self-report: t = -2.38, p = .02). Further, the initial assessment identified 7 out of 30 siblings (23.3 %) with a full and 43.3 % with a partial DSM-IV related PTSD.

Table 7. *Demographic and medical characteristics of the sample (N=30)*

	Intervention (N=14)	Control (N=16)	t*	χ^2**	Z***	p
Age at baseline (years)						
Mean (SD)	9.4 (2.7)	11.0 (2.9)	1.6			.12
Sex						
Boys (%)	9 (64.3)	9 (56.2)				
Girls (%)	5 (35.7)	7 (43.8)		0.20		.65
Socio-economic status						
Lower (%)	0 (0)	0 (0)			-8.46	.40
Middle (%)	10 (71.4)	9 (56.2)				
Upper (%)	4 (28.6)	7 (43.8)				
Mean (SD)	1.71 (0.47)	1.56 (0.51)				
Birth order **						
Younger (%)	4 (28.6)	8 (50)				
Older (%)	10 (71.4)	7 (43.8)		1.83		.18
Type of diagnosis in ill child						
Hematological malignancies (%)	7 (43.8)	10 (71.4)				
Brain or other solid tumor (%)	9 (56.3)	4 (28.6)		2.33		.13
Days hospitalized						
T0, Mean (SD)	23.9 (6.1)	22.8 (9.0)	0.39			.70
T1, Mean (SD)	42.6 (16.5)	40.4 (10.5)	0.43			.67
T2, Mean (SD)	67.9 (19.7)	59.4 (17.9)	1.25			.22

*Independent two-sample t-test ** Chi-square analysis *** Mann-Whitney U test

4.2 Intervention effects

Table 8 presents the results of the repeated measure ANOVA.

Medical knowledge: Results for the medical knowledge scale showed a main time effect (p = .01) with significant increase of knowledge from T1 to T2 (Table 8). No group or group × time effect could be seen in the ANOVA. However, comparing mean knowledge levels of intervention and control groups at T2 revealed significantly better knowledge in the intervention group (M_{int} = 1.62, SD_{int} = 0.36; $M_{control}$ = 1.37, $SD_{control}$ = 0.27; t = 2.20, p = .04). Effect sizes were in the medium range (T2: d = .78). No significant mean differences were found at T0 and T1.

Social support: A significant main effect for group (p = .04) was found for the number of persons providing support, with the intervention group having a higher number of persons.

Time and group × time showed no effect on the social support measure. Effect sizes were in the medium range (T1: d = .61; T2: d = .77).

Health related quality of life, child report: The KIDSCREEN child report total score revealed a significant time effect (p ≦ .001), with improvement of HRQoL from T0 to T1, T1 to T2, and T0 to T2 in both groups. The intervention had no significant influence on the KIDSCREEN total score as reported by siblings. However, the KIDSCREEN self-report subscale "psychological well-being" showed a significant main effect of the intervention (p = .03) as well as a significant time effect (p ≦ .001), indicating improvements over time in both groups, but with siblings in the intervention group demonstrating better psychological well-being as compared to the control group. Effect sizes were large for T1 (d = .99) and rather small for T2 (d = .26). Main effects of the intervention or effects of intervention × time interactions could not be found in any other subscale of the KIDSCREEN. Two more KIDSCREEN subscales in the child version showed significant improvements over time: "Autonomy and parents" (F = 3.95; P = .03) and "school environment" (F = 8.45; p = .001).

Health related quality of life; parent report: For the parent version of the KIDSCREEN no significant effect was noted with respect to group and group × time interaction. But the analyses showed a significant time effect with significant improvement of HRQoL from T1 to T2 and T0 to T2.

Posttraumatic stress symptoms: ANOVA results showed a significant time effect (p = .02) in both groups on the UCLA RI scales. However, the time effect was no longer significant in the post-hoc pairwise comparisons using Bonferroni tests. There was no main effect for group in posttraumatic stress symptoms. Full PTSD diagnosis decreased in the whole sample, with 5 siblings (16.7%) that met full diagnosis criteria at T1 and 3 siblings (10%) at T2.

Anxiety: Siblings' anxiety showed a time effect (p = .02) with a significant reduction of anxiety in both groups from T1 to T2. No group or time × group effect could be found. A look at the baseline T0 scores of anxiety and PTSS shows that the intervention group starts out with a considerably higher level although not statistically significant. We therefore also conducted repeated measures ANCOVAS with T0 scores of anxiety and PTSS as covariates. But still no group effect could be shown.

Table 8. *Means, standard deviations and analysis of variance for repeated measures (ANOVA)*

	Intervention (N=14)	**Control group** (N=16)	**ANOVA F**		
	M (SD)	**M (SD)**	**Time (T)**	**Group (G)**	**T × G**
Medical knowledge			5.11**	2.61	0.27
T0, mean score	1.45 (0.56)	1.23 (0.37)			
T1, mean score	1.46 (0.48)	1.28 (0.35)			
T2, mean score	1.63 (0.36)	1.37 (0.27)			
Social support			0.24	4.58*	0.51
T0, number	10.43 (1.16)	9.75 (1.88)			
T1, number	10.93 (0.83)	9.69 (2.73)			
T2, number	10.86 (0.77)	9.25 (2.86)			
HRQoL: KIDSCREEN child version total score			13.88***	1.83	0.60
T0, mean T-scores	48.87 (4.79)	46.01 (6.75)			
T1, mean T-scores	52.95 (5.01)	49.12 (6.68)			
T2, mean T-scores	53.64 (6.94)	52.09 (7.36)			
HRQoL: KIDSCREEN child version subscale psychological well-being			37.16***	5.29*	1.57
T0, T-scores	39.59 (4.37)	36.69 (4.28)			
T1, T-scores	55.76 (8.45)	47.06 (8.96)			
T2, T-scores	54.76 (10.77)	52.08 (9.81)			
HRQoL: KIDSCREEN parent version total score			4.14*	1.11	1.49
T0, mean T-scores	48.70 (4.97)	48.60 (6.27)			
T1, mean T-scores	49.99 (5.16)	53.52 (6.22)			
T2, mean T-scores	49.20 (4.05)	51.68 (7.81)			
Posttraumatic stress symptoms: UCLA RI			4.15*	0.91	0.86
T0, total score	22.23 (14.06)	15.57 (11.89)			
T1, total score	15.00 (9.79)	13.71 (11.36)			
T2, total score	15.00 (13.04)	11.64 (11.45)			
Anxiety: SCAS total score			5.35*	0.64	1.38
T0, total score	29.14 (25.52)	20.47 (17.15)			
T1, total score	18.43 (12.35)	17.93 (15.62)			
T2, total score	19.71 (12.86)	15.33 (15.63)			

*p ≦ .05; **p ≦ .01; ***p ≦ .001

5. Discussion

The aim of this study was to evaluate the effectiveness of an early psychological intervention with siblings of newly diagnosed pediatric cancer patients. Our findings suggest that a two-session intervention leads to better psychological well-being and increases the amount of social support and amount of medical knowledge. However, there were no intervention effects in measures of proxy-rated HRQoL, PTSS, and anxiety.

Since children often rely on their own interpretations of illness and sometimes have a distorted picture of it, it is crucial to provide age-appropriate medical information (Wilkins & Woodgate, 2005). The results of our study show better knowledge in siblings after the intervention. This beneficial finding is in line with previous intervention studies (Dolgin, Somer, Zaidel, & Zaizov, 1997; Sahler & Carpenter, 1989). Although this study did not examine the effects of knowledge, other researchers found that enhanced knowledge had a positive impact on siblings' adjustment and social competences (Evans et al., 1992; Last & Grootenhuis, 1998).

The number of individuals available for social support increased for siblings who took part in the intervention. This improvement can probably be attributed to the coping part of our intervention, where many of the coping skills discussed involved looking for social support. Moreover, the psycho-educational booklet provided to parents covered social support issues. Although having a higher number of available individuals does not necessarily mean better social support, having a greater number of available potential partners increases chances of receiving helpful social support, especially for our population, which is confronted with decreased social resources in the core family due to the cancer diagnosis (Alderfer et al., 2010). Other studies identified social support as an important construct that may play a critical protective role in the psychosocial adjustment of siblings of cancer patients (Alderfer & Hodges, 2010; Barrera, Fleming, & Khan, 2004). Our study is the first to include social support as an outcome measure in the evaluation of interventions for siblings.

Siblings' self-reports indicated a significant increase in psychological well-being after the intervention. Even though this was the only subscale of the KIDSCREEN questionnaire showing group differences, well-being represents an essential part of siblings' psychological adjustment (positive emotions, satisfaction with life, and balanced emotionality). This positive effect of our intervention on HRQoL is consistent with two other studies (Packman, Chesterman, vanZutphen, Golan, & Amylon, 2004; Packman et al., 2005). Notably, this result was not apparent from the mothers' reports of the child's HRQoL. This might be due to parents' difficulties in judging the child's HRQoL in an emotional domain such as psychological well-being (Eiser & Morse, 2001).

Contrary to our hypothesis, anxiety and PTSS were not improved by the intervention, although trends in the desired directions could be observed. Anxiety was not clinically increased in our sample compared to norms, a finding also seen in previous studies (Alderfer & Hodges, 2010; Dolgin et al., 1997; Houtzager et al., 2004), and might therefore be an

inappropriate measure to assess effects of an intervention. PTSS, on the other hand, was high at baseline, with almost a quarter of our sample qualifying for full diagnosis of PTSD. These findings are in line with previous studies that did not operate with DSM-IV related instruments but revealed similar high numbers of PTSS scores (Alderfer & Hodges, 2010; Alderfer et al., 2003; Packman, 2004; Packman et al., 2004). However, our intervention might have been too unspecific regarding PTSS to help siblings with relevant symptoms.

Alternative explanations for absent intervention effects should also be considered. It is possible that families' involvement in the study and the data collection itself increased and perhaps improved communication between parents and siblings (Williams et al., 2003) and therefore led to better adjustment in both study groups. Other unspecific factors may also play an important role. By enrolling siblings in a specific sibling program, parents may demonstrate their concern for them and may develop particular efforts to spend time with them (Barrera, Chung, & Fleming, 2004). Likewise, we have to consider that our whole population had standard psychological care at hand, and this might have leveled group differences. It is also possible that our age range was too broad; whereas certain age groups could have benefited from the intervention, for others the intervention may not have been appropriate.

This study has a number of limitations. First, the fact that our intervention was for the early time period after diagnosis made recruitment more difficult and resulted in a rather small sample. We may therefore have lacked power to find intervention effects, and we were not able to perform subgroup analyses (age, sex). Second, we developed the measures for social support and medical knowledge ourselves, and they have limited reliability and validity. Future studies should use validated instruments for these outcome measures. Third, families with lower socio-economic status were not represented in the sample. This might be due to the inclusion criterion of fluency in German, which excluded families with an immigrant background.

Despite these limitations, this study has several strengths, including its randomized controlled prospective design and the manualized intervention for the early stage after diagnosis. Moreover, statistical conditions were good, with no socio-demographic differences between study participants and non-participants, no differences between intervention and control groups on all baseline scores, and almost no attrition.

There are also implications for future research. Since our results as well as prior studies showed that siblings of cancer patients most often fall into a normal range of functioning, standardized measures of psychopathology might be unable to capture the unique difficulties of siblings. Broader measures assessing the child's HRQoL or social support may be more sensitive. Also, considering the high rate of siblings with PTSD symptoms, future research on interventions should integrate a more trauma-focused approach.

The early, two-session individual psychological intervention evaluated here is promising for improving some aspects for healthy siblings. Efforts to provide medical information, enhance coping skills, and inform parents showed no beneficial effects on anxiety, PTSS, and parent-reported HRQoL but seemed to be rewarded by increased child-reported well-being, social support, and medical knowledge.

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V. General Discussion

The primary aims of our project were to develop, implement and evaluate a individual, standardized, early psychological intervention for school-aged siblings of pediatric cancer patients. In accordance with current methodological standards in intervention research, a randomized controlled trial was used for the evaluation of the sibling intervention.

As a first step towards developing our psychological intervention, we reviewed the research on existing psychological interventions for our target group. The results of this review, from which we drew information that was helpful towards determining the design and content of our intervention, were presented in Chapter II. As a second step, we sought to learn more about the specific situation of siblings in the immediate aftermath of cancer being diagnosed, by conducting a qualitative pre-study. In this survey, seven siblings of pediatric cancer patients were interviewed about their experiences in several areas of life. Qualitative content analysis was used to extract relevant issues from these interviews. This part of our project was presented in Chapter III. Findings from both, the review and the qualitative sibling interviews, were used to develop our sibling intervention (Chapter IV). This early individual psychological intervention then was tested empirically by means of a randomized controlled trial. In this concluding chapter, the main findings of the last three chapters will be summarized and several broader issues that arise from these findings will be discussed. Also, limitations of the project will be considered; and suggestions for clinical practice, as well as recommendations for future research, will be provided.

1. Summary of findings

The main findings of the studies are summarized in Table 9 and will be presented briefly below.

During our review, we discovered that evaluated interventions targeting siblings of pediatric cancer patients remain rare and of relatively low scientific merit. Interventions in a group or camp format are overrepresented and most interventions focus upon enhancing coping strategies and providing siblings with medical knowledge. According to our review, there is tentative evidence that psychological interventions with siblings can reduce symptoms of depression, and enhance HRQoL as well as medical knowledge.

As a result of our qualitative survey, in which siblings were asked about their experiences in the first half-year after the diagnosis of cancer in their sibling, we identified 22 categories of experience covering all areas of life (hospital, school, family, peers, and the ill child). Siblings often reported difficulties, like the absence of parents, dealing with the ill child's or other patients' suffering and appearance, or impaired school achievement. On the other hand, some developed helpful coping strategies to encounter those stressors. Also, important resources were mentioned, such as peer relationships, and gains like increased family cohesion.

Finally, as a result of our own two-session psychological intervention that focused on medical information, coping, and psycho-education, siblings at follow-up reported better social support, increased psychological well-being, and advanced medical knowledge relative to untreated controls. However, we failed to detect significant group differences at follow-up concerning parent-rated HRQoL, PTS, anxiety, depression, behavioral problems, perception of illness, or the number of coping strategies. Results of measures not considered in the paper are presented in Table 10 in the Appendix.

Table 9. *Summary of main findings of all three studies*

Authors / Content	Sample	Method	Main findings
Prchal & Landolt, 2009 Review of sibling interventions *Chapter II*	14 studies (10 published articles / 4 dissertations) representing 11 sibling interventions	Descriptive review, applying effect sizes when possible	• Mostly pre/post-intervention designs, only 3 RCTs • 10 of 11 intervention with group/camp format, only one individual intervention • Outcome assessment: improvements in depression, negative mood, medical knowledge, HRQoL • Outcome assessment: inconsistent findings regarding anxiety, behavioral problems, PTS, social adjustment, self-esteem, perception of illness
Prchal & Landolt, 2011 Qualitative sibling interviews *Chapter III*	7 siblings	Semi-structured interview content analysis	• 22 categories of siblings' experiences in all 5 domains (hospital, school, peers, family life, ill child) • Main difficulties: absence of parents, dealing with the ill child's or other patients' suffering and appearance, impaired school achievement • Resources and gains: Peer relationship, positive encounters with hospital staff and other impatiens, increased family cohesion • Helpful coping strategies: Active involvement in care of the ill child, getting to know the hospital, strategies in dealing with questions from others
Prchal, Graf, Bergsträsser & Landolt, 2011 Intervention evaluation *Chapter IV*	30 siblings (15/15)	Intervention evaluation Randomized controlled trial	• At baseline siblings showed anxiety, depression, problem behavior and proxy-reported HRQoL within the normal range • At baseline self-reported HRQoL was lower compared to norm, 23.3 % of siblings showed full and 43.3 % partial PTSD • Outcome assessment: significant increase in social support, self-reported HRQOL subscale "psychological wellbeing", medical knowledge • Outcome assessment: no effect on proxy-reported HRQoL, PTS, anxiety, depression, behavioral problems, number of coping strategies

2. Reflections on the findings

2.1 The present study in the field of evaluated sibling interventions

Our review demonstrated that the number of evaluated sibling interventions remains small and that the existing reports are mostly methodologically weak. Considering that two of the three RCTs included in our review remain unpublished dissertations (Atherton, 1984; Cimini, 1986) and that the final RCT evaluated a family therapy approach in just a small number of siblings (Kazak et al., 2004), the state of evidence seems even weaker. Furthermore, the majority of evaluated sibling interventions were designed for use in a group or camp format. This could mean that interventions designed for individual siblings are rare. Another possible explanation, however, is that group or camp formats are more likely to be evaluated, since it is easier and more time-efficient to collect data from a group than multiple individuals treated separately. A recently-published paper describing child life services provided to siblings in the United States and Canada (Newton, Wolgemuth, Gallivan, & Wrightson, 2010) reported that sibling support services are available at about half of children's hospitals. However, a vast majority (72%) of respondents (administrative and clinical practice leads) indicated that these services were not evaluated in terms of improved outcomes. Even though this study failed to systematically differentiate between individual and group formats, it becomes clear that quite a few of the above-mentioned services were provided at the individual versus group level (e.g., therapeutic play, palliative care support). Therefore, we assume that individual sibling support is something that exists at many institutions, but that such services rarely are formally evaluated. Our study was intended to fill an essential gap in the field of sibling intervention studies, by developing an individual sibling intervention and examining its effects by means of a randomized controlled trial.

Our study had several other methodological advantages over previous intervention studies. First, we incorporated time since diagnosis in our intervention procedure. Whereas most sibling intervention studies included in our review either used broad inclusion criteria concerning the time lag since the diagnosis of cancer or failed to take the time since diagnosis into consideration at all, our intervention was provided at an early stage, within the first two months of diagnosis. Thus, the intervention could be tailored to the needs and stressors associated with this particular stage of treatment. Second, the intervention was standardized and well-documented to allow for study replication, details that were neglected in most previously-reported studies. Third, effect sizes were calculated to determine whether significant mean differences were detectable. And finally, all necessary data for the further calculation of effect sizes were provided, thereby allowing for the comparison of different types of intervention in future meta-analyses.

The outcome results of the main study are mostly in line with the findings of our review. The medical knowledge and HRQoL of the siblings in the intervention group improved significantly as a result of our intervention, a finding consistent with previous intervention studies. Findings related to anxiety variables, problem behaviors and PTS were inconsistent in our review, and we were unable to detect any significant effects of our intervention on

these variables either. Regarding depression scores, however, the findings of our intervention evaluation do not correspond with previous publications. Contrary to the four previously-published studies that assessed depression in siblings and uniformly identified improvements in depressive symptoms post-intervention, we identified no significant effects of our intervention on the depression score. This comparison must be interpreted with caution, however. To begin with, the effect sizes for depression in previous studies were small. Secondly, three of the four studies reporting on depression utilized the same intervention program. Finally, that we could not identify any significant improvement in depression also might have to do with the depression rates of our participant siblings at baseline, which were already low and within the normal range.
Social support was not an outcome category in previous studies. Therefore, any comparisons of our findings versus the literature are not possible.

2.2 The influence of the qualitative data on the intervention content

Results from our qualitative study influenced our sibling intervention in several ways. Due to the fact that sibling distress was apparent in all areas of life, we placed emphasis on asking siblings during the intervention not only about stressors in the family or relating to the ill child, but also stressors explicitly related to their peers and their school, and stressors experienced within the hospital. The descriptions collected in the qualitative study, of important experiences and helpful coping strategies that siblings managed to utilize, yielded a list of topics that was available during the coping part of our intervention. This list was presented to siblings and facilitated constructive discussions about personal stressors and effective coping strategies. Being able to name difficulties experienced by other siblings gave participants in the intervention group a more global picture of their situation and helped to normalize their feelings.

2.3 Posttraumatic stress in siblings of pediatric cancer patients

Given that mean scores of psychopathological measures like anxiety or depression are often within the normal range in cancer siblings, there is a need to carefully identify which outcome measures should be targeted and assessed by sibling interventions. Posttraumatic stress might be an important variable to measure while working with siblings of pediatric cancer patients.
To conceptualize the ongoing cancer-related distress of pediatric cancer patients and their parents, a posttraumatic stress model has been used effectively as a framework by (Hobbie et al., 2000; Kazak et al., 1997). Although siblings of childhood cancer might not have the same level of exposure to stressors as parents and patients themselves, siblings still experience traumatic aspects of the cancer. Results from (Alderfer, Labay, & Kazak, 2003) as well as the findings from our own qualitative study show that siblings are often exposed to the physical and/or emotional suffering of their ill brother or sister; they might experience fearful situations in the hospital with other patients; and many are extremely fearful that the ill child

will die. In addition to this, they experience these stressful situations at a time when parental support for them often is limited. These perceptions may be traumatic and might explain why siblings are likely to show symptoms of PTS. Results of our intervention study (Chapter IV) as well as earlier reports assessing PTS, display consistently high PTS rates in this population (Alderfer & Hodges, 2010; Alderfer et al., 2003; Packman, 2004). It therefore seems necessary that intervention studies assess PTS as an outcome measure, and that interventions themselves contain trauma-centered therapeutic elements, an approach that has not yet been implemented for siblings specifically. Prior to our study, only two published interventional studies assessed PTS as an outcome variable (Kazak et al., 2004; Packman, Chesterman, vanZutphen, Golan, & Amylon, 2004).

2.4 The role of social support for cancer siblings

The present intervention study is, to our knowledge, the first to include social support measures in the evaluation of a sibling intervention. Outcome assessment revealed significant improvements in the amount of social support siblings received after the intervention. This finding suggests that the intervention part that concentrated on enhancing coping skills, especially social support seeking, and the attempt to address social support issues in the booklet for parents were successful. Further, social support was identified in other studies as an important construct that may play a critical role in the psychosocial adjustment of siblings (Alderfer & Hodges, 2010; Barrera, Fleming, & Khan, 2004). Therefore, the results of our study, along with other recent research, indicate that a sibling's social support needs to be carefully evaluated and sibling interventions should incorporate its enhancement.

The family is the primary social support system for children and adolescents, and greater family support has consistently been associated with less psychological distress and behavioral adjustment (Newman, Newman, Griffen, O'Connor, & Spas, 2007); but, due to inherent changes in the family after cancer is diagnosed in one child, an important source of social support for siblings is impaired or not sufficiently available. Hence, in addition to the core family, other support systems, like peers and school, should be activated by interventions. Our qualitative study revealed that peers are a particularly important source of support. Supportive relationships with peers were detected as resources that help siblings to cope with their situation. Peers allow siblings of cancer patients to talk about their situation, to enjoy distraction, and to experience normal life away from serious illness in the family. As mentioned by Alderfer and Hodges (2010), school-based social support, especially from teachers, also seems helpful in creating a warm and reassuring environment, leading to better adjustment in siblings. There is also the fact that teachers who are appropriately educated about cancer may dispel other's misconceptions and may effectively answer any medical questions that arise in school, questions that the sibling does not need to answer anymore. Accordingly, giving siblings the opportunity to spend time with their peers and inform their school should be an important goal of any psychological intervention with cancer siblings.

3. Critical review and limitations of the project

The current intervention study has several limitations. Thirty siblings participated in the intervention study, a rather small number for a randomized controlled design, resulting in insufficient statistical power. Due to the relatively small pediatric oncology population in any institution and the fact that not all children with cancer have siblings, limited sample size is a frequent methodological problem (Patenaude & Kupst, 2005). The issue of small sample size also was apparent in our review of the literature, as well as in a recently-published review on interventions with siblings of children with chronic illness or disability (Hartling et al., 2010).

This methodological weakness hampered our initial plan to examine the impact of disease and family variables on adjustment and intervention effect. This is unfortunate, because our clinical impression revealed large differences between families regarding family resources and approaches to support siblings. These variables might explain a large part of sibling adjustment and could substantially influence the effectiveness of an intervention.

One reason for the small sample size in our project was recruitment difficulties. Attendance in our study was moderate, with one third of the siblings who met the inclusion criteria not participating (response rate 66.7%). Due to our intent for early sibling intervention, recruitment of families had to take place within the first few weeks after the cancer diagnosis. This initial phase is among the most stressful periods parents encounter, and many reported that they were overwhelmed by the amount of information they received (McCubbin, Balling, Possin, S, & Bryne, 2002). Our participants were underage siblings, so we had to inform their parents about the study and to obtain consent from them. It is possible that the parents were overwhelmed by our request and that families who refused to participate in our study did not want any additional burden placed upon them during that early stage after diagnosis. In this way, difficulties in recruitment increased the risk of selection bias. For example, it is possible that families who volunteer for research studies tend to be functioning well, relative to those who choose not to participate, and this may compromise the external validity of a study's findings. Kazak et al. (2003) reported that families at the highest psychosocial risk at diagnosis were less likely to complete subsequent data collection. Therefore, we cannot rule out the possibility that families and siblings who participated in our study represented a rather resilient group, and that those at greater risk for maladjustment we not surveyed. Moreover, families with a lower socio-economic background were not represented in the sample, probably due to the criterion of fluency in German. This criterion excluded at least some families with an immigrant background.

The design of our intervention study did not allow us to determine the effectiveness of single components of the intervention. Further, our attempt to at least qualitatively explore satisfaction with certain elements of the intervention failed. Questions about how helpful parents and siblings considered particular parts of the interventions were implemented in the follow-up assessment at T2. However, roughly half way through recruitment, we realized that a significant subgroup of parents and siblings were unable to state whether they had been in the

control or intervention group half a year after the first interview. One reason for this could have been that the information we had provided them had been ambiguous. With the intent to avoid research bias, the interviewer at T0 was not informed to which group, intervention versus control, a particular family had been allocated. Only directly after the interview and immediately before the intervention started was group allocation apparent. Thus, some families might not have realized whether the intervention was part of the interview or not. Later on, we emphasized informing families clearly about their assignment in the study, but it was too late for us to evaluate the data derived from the satisfaction questionnaire.

Considering the broad age group of participating siblings (6 to 17 years), we also cannot ensure that our approach was age-appropriate for all participants. Although we took care to use age-appropriate language and chose different visual aids according to a child's developmental stage, the main part of the intervention was purely verbal. This might have been too difficult for younger children. Further, especially during the first session, which also included the baseline interview, the attention span required of participating children was quite long, as we combined the first interview with the first intervention session for economic reasons. This could have disadvantaged siblings with lesser abilities to concentrate.

4. Directions for future research and clinical implications

Siblings' experiences are quite unique and it would be worth conducting further investigations to clarify them. Future studies also should include sample sizes that ensure sufficient statistical power to detect even small differences in outcomes, to control for the moderating role of illness or environmental variables, and to enhance external validity. Multi-site studies are often recommended as essential to recruiting samples of adequate size.

Most sibling interventions are complex and have multiple components; but study designs rarely allow investigators to identify the most effective components of a given intervention. To determine the effectiveness of particular elements of interventions, future research should engage in process evaluation (see Kelly, Nixon, & Bickman, 2000).

Measurements in pediatric psycho-oncology research have progressed from clinical impressions and case studies to the use of standardized measures of anxiety, depression, and behavioral adjustment to determine the prevalence of these outcomes (Patenaude & Kupst, 2005). These measures have the advantage of being based upon normalized data and having good psychometric properties. On the other hand, standardized quantitative measures of psychopathology might not be able to capture the unique difficulties of the siblings of children with cancer (Long & Marsland, 2011). Future researchers must take into consideration that difficulties in sibling adjustment may not fit into current categories of psychopathology, so that the range of outcome variables should be expanded. Although previously-published studies have identified subsets of siblings who meet the criteria for anxiety and depression, and our results revealed a high incidence of cancer-related PTS, future research should pay more attention to the impact of cancer on social, academic and somatic domains, and on health-related quality of life.

Since social support was found to be an important variable in sibling adjustment, it seems warranted to expand future sibling interventions in terms of the facilitation of social support, in particular from peers and school personnel. Considering the high rate of siblings with PTS symptoms in our study, the integration of a trauma-focused approach in future interventions is likewise recommended.

The findings of the current study provide a rich foundation from which health care professionals can build a better understanding of sibling adjustments to pediatric cancer, especially in the first months after the diagnosis of cancer is made; and this should help them to provide more appropriate care. Our findings clearly demonstrate that a sibling's distress is substantial in the first months after the diagnosis of cancer is made in a child. Health care professionals should ensure that information and support is available for siblings early on, and should call parents' attention to all siblings' needs and possible reactions. Since siblings experience stressful situations not only in the hospital, but also in their daily life at home and in school, these situations should be addressed directly. Social support from school and peers should be recognized as an important resource worth bolstering. Finally, professionals must keep in mind that siblings of cancer patients have an increased risk of developing PTS symptoms. It is unrealistic to assume that the stressors siblings face when cancer occurs in the family can be entirely prevented; but it is reasonable to expect that, by providing appropriate early psychological and social support, these stressors can be addressed and successfully managed before long-term psychological maladjustment emerges.

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Appendix

Appendix A: Psychoeducational brochure for parents
Appendix B: Additional ANOVA results

Aktivitäten ausserhalb der Familie fördern

Ermutigen Sie das gesunde Geschwister Dinge zu unternehmen, die ihm Spass machen. Dazu gehören Hobbys nach der Schule und Kontakte mit Gleichaltrigen. Vermitteln Sie dem Kind, das es in Ordnung ist, wenn es Freude an Aktivitäten ausserhalb des familiären Rahmens und fernab von all den Sorgen rund um die Krankheit hat.

Kontakt

Kinderspital Zürich
Universitäts-Kinderkliniken
Steinwiesstrasse 75
CH-8032 Zürich

Telefon Zentrale 01 266 71 11
Telefon PD Dr. Landolt 01 266 73 96
(Fachpsychologe FSP)

Den Kindern alles Gute.

Geschwister krebskranker Kinder

KINDERSPITAL ZÜRICH
Universitäts-Kinderkliniken · Eleonoren-Stiftung

Aktivitäten ausserhalb der Familie fördern

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Universitäts-Kinderkliniken · Eleonoren-Stiftung

Liebe Eltern,

eines Ihrer Kinder ist seit kurzer Zeit wegen einer onkologischen Erkrankung bei uns im Spital in Behandlung. Die Erkrankung eines Kindes bringt den Rhythmus der ganzen Familie durcheinander. Von einem Tag auf den anderen musste Ihr Kind ins Spital. Sie als Eltern sind körperlich und psychisch mit dem kranken Kind so sehr beschäftigt, dass Sie kaum mehr Zeit und Kraft für etwas anderes haben. Es ist klar, dass das kranke Kind im Mittelpunkt Ihres Handelns und Ihrer Gedanken steht. Den Geschwistern von krebskranken Kindern kommt eine besondere Rolle zu. Sie müssen in dieser Zeit ihre Bedürfnisse oft zurückstecken, mehr Verantwortung übernehmen und mehr Zeit ohne ihre nächsten Familienmitglieder verbringen. Nicht selten plagen die Geschwister Gefühle von Eifersucht, Angst oder Schuld. Es gibt unausgesprochene Fragen und einige Geschwister haben den Wunsch, ihre Eltern zu schonen.

Jede Familiensituation ist anders und die Belastung der Geschwisterkinder äussert sich in unterschiedlicher Weise. Dennoch gibt es einige typische Reaktionen von Geschwistern in dieser aussergewöhnlichen Situation. Die vorliegende Broschüre stellt im ersten Teil verschiedene Situationen und Lebensbereiche vor, in welchen die Krebserkrankung das gesunde Geschwisterkind stark betrifft. Im zweiten Teil werden Möglichkeiten zur Unterstützung der Geschwister aufgezeigt. Wir hoffen, dass einige Anregungen und Hilfestellungen Ihrer Familie weiterhelfen können.

Die Situation der Geschwister

Diagnose

Wie alle Familienmitglieder erleben auch die Geschwister die Diagnose Krebs als grossen Schock. Oft bringen Sie die Krankheit automatisch mit einem tödlichen Ausgang in Verbindung, denn häufig kennen sie Erwachsene, die an Krebs gestorben sind, oder sie haben schon von solchen Schicksalen gehört. Weil sie ihr Wissen über Krebs von Erfahrungen mit älteren Menschen ableiten, steht die Angst vor einer schlechten Prognose und einem tödlichen Verlauf im Vordergrund. Viele wissen nicht, dass die Heilungschancen in Kindesalter oft ausgesprochen gut sind und reagieren daher mit Trauer und Angst wegen des möglichen Verlustes des Geschwisters. Manche Geschwister entwickeln auch grosse Wut darüber, dass ausgerechnet ihr Bruder oder ihre Schwester betroffen ist.

Familienleben

In der ersten Zeit nach der Diagnose muss sich Ihre Familie neu organisieren und orientieren. Sie als Eltern sind häufig im Krankenhaus und wollen sich intensiv um das kranke Kind kümmern. Manchmal werden Verwandte oder Nachbarn für die Versorgung der Familie hinzugezogen, das gesunde Geschwister muss sich an neue Personen und Umgebungen gewöhnen, und, je nach Alter, auch zusätzliche Verantwortung übernehmen. So bringt es die Situation mit sich, dass die gesunden Geschwisterkinder im Alltag oft zu kurz kommen, ohne dass dies beabsichtigt ist. Hinzu kommt, dass die Kinder Sie als Eltern in dieser Zeit in einer Situation erleben, der Sie häufig ängstlich und hilflos gegenüberstehen. Dies kann insbesondere jüngere Kinder verunsichern. Sie müssen feststellen, dass ihre Eltern sie nicht uneingeschränkt vor Gefahren schützen können.

Jugendliche zeigen sich im Hinblick auf das veränderte Familienleben und die Übernahme von Verantwortung oft verständnisvoll. Sie nehmen Rücksicht, verzichten bewusst auf Aufmerksamkeit der Eltern und übernehmen freiwillig zusätzliche Aufgaben. Auf emotionaler Ebene kann es aber sein, dass sie sich, ohne es zuzugeben, dennoch zurückgestellt fühlen. Jüngere Kinder reagieren in der Regel offener und direkter, zeigen ihre Enttäuschung und sagen, dass sie sich benachteiligt fühlen.

Schule

Insbesondere in der ersten Zeit nach der Diagnose können die Sorge um das kranke Kind und die Veränderungen in der Familie dazu führen, dass das Geschwister sich in der Schule schlecht konzentrieren kann, oder dass es müde und lustlos ist. Es kann vorkommen, dass sich die Schulleistungen unter diesen Umständen verschlechtern. Oftmals erachten Kinder wie auch Eltern die Schule angesichts der Konfrontation mit einer existentiellen Bedrohung durch die Krankheit als nicht mehr so wichtig. Schlechtere Schulleistungen können auch damit zu tun haben, dass die Eltern ihr Kind bei Hausaufgaben und Prüfungsvorbereitungen weniger unterstützen können.

Beziehung zum kranken Geschwister

Aufgrund der häufigen Spitalaufenthalte des erkrankten Kindes sehen sich die Geschwister seltener. Obwohl sie für das kranke Geschwister Mitleid empfinden, können sie ihm gegenüber durchaus auch ärgerlich oder wütend reagieren. Wie sich die Beziehung der Geschwister untereinander entwickelt, hängt auch vom Alter ab. Mit zunehmendem Alter haben die Kinder ein besseres Verständnis für die Situation und ihre Folgen. Besteht beim gesunden Geschwister jedoch ein Gefühl der Vernachlässigung und die Sorge, weniger geliebt zu werden, kann es zu heftigen Spannungen und Eifersucht zwischen den Geschwistern kommen. Das kranke Kind bekommt viel mehr Aufmerksamkeit und die gesunden Geschwisterkinder erleben, wie das kranke Kind mit Geschenken überhäuft wird und die Eltern weniger streng mit ihnen sind. Gleichzeitig wird vom gesunden Kind mehr Rücksichtsnahme und Verständnis gefordert, was in Anbetracht der Dauer der Erkrankung zur Überforderung werden kann. Vor allem jüngere Kinder entwickeln manchmal die Vorstellung, dass krank sein eine Garantie dafür sei, von den Eltern geliebt und beachtet zu werden. Infolgedessen klagen sie vermehrt über körperliche Beschwerden, für die es keinen medizinischen Befund gibt, die es aber dennoch zu beachten gilt.

Möglichkeiten, Geschwisterkinder zu unterstützen

Sie sind als Eltern in der ersten Zeit nach der Krebsdiagnose enorm gefordert. Die lebensbedrohliche Erkrankung, die eigene psychische Belastung und die Alltagssorgen nehmen grossen Raum ein. Es ist ganz natürlich, wenn die Gefühle und Sorgen sich in erster Linie auf das erkrankte Kind richten und Sie ihre ganze Kraft für seine Betreuung einsetzen. Für anderes bleibt in dieser Zeit wenig Energie und es ist klar, dass den Geschwistern zunächst weniger Aufmerksamkeit und Zuwendung zukommen kann. Dies zu merken, kann bei Eltern Schuldgefühle auslösen. In dieser Situation ist es jedoch wichtig zu erkennen, dass man nicht zu jeder Zeit den Bedürfnissen aller Familienmitglieder gerecht werden kann. Dennoch gibt es einige Hinweise, die in dieser schwierigen Situation hilfreich sein können, vor allem auch im Blick auf die lange Dauer der Krebsbehandlung.

Über die Krankheit sprechen

Geschwister sollen ehrlich und ihrem Alter entsprechend über die Krankheit und die Behandlung informiert werden und Gelegenheit haben, Fragen zu stellen. Es ist wichtig, dass das Geschwisterkind eine genaue Vorstellung davon bekommt, was mit dem Bruder oder der Schwester passiert. Kinder haben zuweilen eigene Phantasien, die oftmals bedrohlicher sind als die Realität. Bringen Sie die Geschwister in Bezug auf die medizinische Situation immer auf den neusten Stand und ermutigen Sie sie, Fragen zu stellen. Erklären Sie dem Kind, dass Krebs eine ernsthafte Erkrankung ist, dass die Ärzte aber alles dafür tun, dass es der Schwester oder dem Bruder wieder besser geht. Stellen Sie klar, dass niemand Schuld daran hat, dass das Kind erkrankt ist, und erwähnen Sie ausdrücklich, dass Krebs nicht ansteckend ist.

Über Gefühle und Sorgen sprechen

Ermutigen Sie Ihr Kind, über seine Sorgen zu sprechen und seine Gefühle auszudrücken. Versichern Sie ihm, dass auch negative Gefühle normal und in Ordnung sind. Es kann vorkommen, dass Ihre Kinder mitbekommen, dass Sie als Eltern traurig sind oder weinen müssen. Es ist in dieser Situation wichtig, dass Sie dem Kind erklären, weshalb Sie traurig sind, damit es nicht denkt, es habe etwas falsch gemacht. Wenn Sie Fragen nicht oder nicht ausreichend beantworten können, geben Sie dies auch zu.

Zeit mit dem Geschwisterkind verbringen

Wenn irgendwie möglich, sollten Sie versuchen, sich ab und zu ausschliesslich mit dem gesunden Geschwisterkind zu

befassen. Versuchen Sie, einen festen Zeitpunkt einzurichten, während dem andere Personen das kranke Kind betreuen und Sie sich ganz dem gesunden Geschwister widmen können. Planen Sie bewusst „Extras“ für das Geschwister ein. Das können Aktivitäten oder Dinge sein, bei denen Sie sich Zeit nehmen, um auf die Bedürfnisse und Wünsche des Geschwisters einzugehen. Unternehmen Sie gemeinsam etwas, was Ihnen und dem Kind Spass macht. Auch nur kurze gemeinsame, aber exklusive Zeit kann sehr hilfreich sein. Fragen Sie das gesunde Geschwisterkind, wie es ihm geht und was sich in seinem Leben ausserhalb der Familie tut.

Geschwister ins Spital mitnehmen

Geben Sie dem Geschwister die Möglichkeit, ins Spital mitzukommen und sich eine Vorstellung von dem Ort und der Behandlung zu machen. Stellen Sie es auf der Abteilung mit Vornamen vor und machen Sie es mit den Räumlichkeiten vertraut. Geschwister brauchen diese konkrete Erfahrung, damit Sie sich keine falschen Vorstellungen vom Spital machen. Erklären Sie dem Geschwister immer, was im Spital mit dem Bruder oder der Schwester geschieht und lassen Sie es auf diese Weise am Geschehen teilhaben. Regelmässig werden im Spital auch Informationsnachmittage speziell für Geschwister von Patienten der Onkologie durchgeführt.

Miteinbeziehen, sich helfen lassen

Versuchen Sie das gesunde Geschwister nicht auszuschliessen, sondern es aktiv in die Betreuung des kranken Kindes einzubeziehen. Fragen Sie das Kind, ob es helfen möchte und geben Sie ihm altersgemässe Aufgaben in der Versorgung des kranken Kindes. Dies können kleine Verrichtungen sein wie z.B. für Getränke sorgen, Dinge bereitstellen, eine Geschichte vorlesen oder Spiele aussuchen und mitnehmen. Wenn das Kind gewisse Aufgaben übernehmen kann, gibt es ihm das Gefühl, in dieser schwierigen Zeit geholfen und etwas Positives beigetragen zu haben.

Annerkennung für Einsatz zeigen

Loben und anerkennen Sie den Einsatz und die Leistungen, die das gesunde Geschwister gerade auch im Zusammenhang mit der ganzen Umstellung im Familienleben und der Betreuung des kranken Kindes vollbringt.

Es ist häufig so, dass vor allem das kranke Kind von Freunden und Verwandten viele Geschenke erhält und im Zentrum der Aufmerksamkeit steht. Wenn Sie den Eindruck haben, dass die Zuwendung allzu einseitig ausfällt, scheuen Sie sich nicht, Ihre Umgebung darauf hinzuweisen.

Klare Abläufe

Versuchen Sie die Terminpläne klar und einheitlich zu halten, so dass das Geschwisterkind im Voraus weiss, von wem es von der Schule abgeholt wird oder wo es über Mittag isst. Halten Sie fest, wo und unter welcher Nummer Sie als Eltern telefonisch erreichbar sind, so dass eine Kontaktaufnahme möglichst jederzeit sichergestellt ist.

Kontakt mit Schule und Lehrer aufnehmen

Informieren Sie die Lehrpersonen der Geschwister, damit diese auf ihre besondere Situation im Schulalltag eingehen, ihnen helfen und Sie stützend begleiten können. Bitten Sie die Lehrkräfte, Ihnen mitzuteilen, falls sie beim Geschwisterkind Veränderungen im Verhalten beobachten.

Fragen von Dritten besprechen

Geschwister werden in der Schule oder bei anderen Gelegenheiten von ihren Freunden, Schulkameraden oder Erwachsenen aus dem Umfeld auf die Krankheit der Schwester oder des Bruders und die aktuelle gesundheitliche Situation angesprochen. Dies kann zu einer grossen Belastung für das Geschwisterkind werden. Ermutigen Sie das Kind dazu, Fragende an die Eltern zu verweisen, wenn es nicht in Stimmung ist, Auskunft zu geben, oder besprechen Sie mit ihrem Kind, wie es auf solche Fragen reagieren kann. Vielleicht können Sie als Eltern auftretende Fragen oder Kommentare vorwegnehmen und mögliche Antworten mit Ihrem Kind schon vorher besprechen. Familien mit entsprechenden technischen Kenntnissen können sich auch überlegen, ob Sie eine Familienhomepage mit Informationen über die Krebserkrankung und die aktuelle Situation der Familie einrichten möchten.

Table 10. *Means, standard deviations and analysis of variance for repeated measures (ANOVA) for behavior problems, depression, perception of illness and coping*

	Intervention (N=14)	**Control group** (N=16)	**ANOVA F**		
	M (SD)	**M (SD)**	**Time (T)**	**Group (G)**	**T × G**
Behavior problems: SDQ			3.78*	0.39	2.97
T0, total score	8.80 (5.36)	9.12 (7.23)			
T1, total score	8.93 (4.53)	6.69 (5.89)			
T2, total score	10.50 (5.19)	8.31 (7.28)			
Depression: DIKJ			4.71**	0.05	0.34
T0, mean score	11.61 (96.01)	12.07 (8.20)			
T1, mean score	8.54 (5.68)	9.97 (5.68)			
T2, mean score	9.31 (6.65)	9.07 (7.94)			
Perception of illness (SPQ)			18.1***	0.11	0.01
T0, total score	1.61 (0.57)	1.56 (0.37)			
T1, total score	1.45 (0.42)	1.44 (0.35)			
T2, total score	1.42 (0.56)	1.35 (0.48)			
HRQoL: KIDSCREEN child version subscale psychological well-being			37.16***	5.29*	1.57
T0, T-scores	39.59 (4.37)	36.69 (4.28)			
T1, T-scores	55.76 (8.45)	47.06 (8.96)			
T2, T-scores	54.76 (10.77)	52.08 (9.81)			
Coping: KIDCOPE			2.41	0.02	0.29
T0, numbers of strat.	3.77 (2.05)	4.19 (2.01)			
T1, numbers of strat.	3.46 (2.15)	3.31 (1.96)			
T2, numbers of strat.	3.15 (2.34)	3.13 (2.00)			

*p ≦ .05; **p ≦ .01; ***p ≦ .001

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Publications

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